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THE IMPACT OF ILLNESS PERCEPTIONS ON HEALTH OUTCOMES IN PATIENTS WITH MULTISYSTEM DISEASES

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Success is not achieved by winning all the time.
Real success comes when we rise after we fall.

Muhammad Ali

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ABBREVIATIONS

Brief-IPQ	Brief Illness Perception Questionnaire
BSSC	Belgian Systemic Sclerosis Cohort
CBT	Cognitive Behavioral Therapy
CFI	Comparative Fit Index
CHD	Coronary Heart Disease
CI	Confidence Interval
COPD	Chronic Obstructive Pulmonary Disease
CSM	Common-Sense Model
dcSSc	diffuse cutaneous Systemic Sclerosis
EUSTAR	European Scleroderma Trials and Research group
EQ-5D-5L	EuroQol five-dimensions with five response levels
EQ-5D VAS	EuroQol Visual Analogue Scale
$FEV_{1\%}$	percent predicted forced expiratory volume in the first second
FIML	Full Information Maximum Likelihood
GP	General Practitioner
HADS	Hospital Anxiety and Depression Scale
HP	Healthcare Professional
ICC	Intraclass Correlation Coefficient
I-CVI	Content Validity Index at item level
IPQ	Illness Perception Questionnaire;
IPQ-R	revised Illness Perception Questionnaire
IPQ-R HP	revised Illness Perception Questionnaire for Healthcare Professionals
k*	modified kappa index
lcSSc	limited cutaneous Systemic Sclerosis
MLR	Maximum Likelihood Estimation with robust standard errors
MOOSE	Meta-analysis Of Observational Studies in Epidemiology
PASI	Psoriasis Area and Severity Index
p_c	probability of chance occurrence
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO	International Prospective Register of Systematic Reviews
RMSEA	Root Mean Square Error of Approximation
SBS χ^2	Satorra-Bentler scaled chi-square statistic
S-CVI	Overall Scale Content Validity Index
$S\text{-}CVI_{Ave}$	Overall Scale Content Validity Index average
SD	Standard Deviation
SELENA	Safety of Estrogens in Lupus Erythematosus National Assessment
SLE	Systemic Lupus Erythematosus
SLEDAI	Systemic Lupus Erythematosus Disease Activity Index
SSc	Systemic Sclerosis

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GENERAL INTRODUCTION

partly based on Seher Arat, Joris Vandenberghe, Philip Moons, René Westhovens

Patients' perceptions of their rheumatic condition: why does it matter and how can healthcare professionals influence or deal with these perceptions?

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1. What are illness perceptions?

Illness perceptions are as old as mankind. Long before the term was formulated by researchers, people have tried to interpret symptoms and illnesses that they and those close to them encountered and thus give meaning to their experiences [1]. 'Illness perceptions' is a term used to refer to the mental representations and personal ideas that people have about an illness, which is a dynamic process and can fluctuate over time [2]. These personal ideas about illness can be seen in the broader context of the self and the sociocultural system. This means that they are not merely states of the individual, experienced and interpreted at the physical and psychological levels but that they are also shaped by the pre-illness self, the surrounding culture, institutions, and social networks [3].

Currently, illness perceptions are seen as frameworks or mental models that individuals construct to make sense of their symptoms and medical conditions [4]. These illness models are implicit and can be very specific to the individual. Also, patients with the same condition can have different perceptions regarding their illness [5,6]. Nevertheless, consistent patterns can be identified in the way individuals generate illness perceptions. These perceptions are an important key to understanding individuals' illness behavior, no less important than the classical so-called objective indicators of the disease. The concept of illness perceptions has been introduced in the early 1980s by Howard Leventhal and colleagues [7,8] in the Common-Sense Model of Self-Regulation in health and illness. The work performed and discussed in this PhD thesis is based on this model.

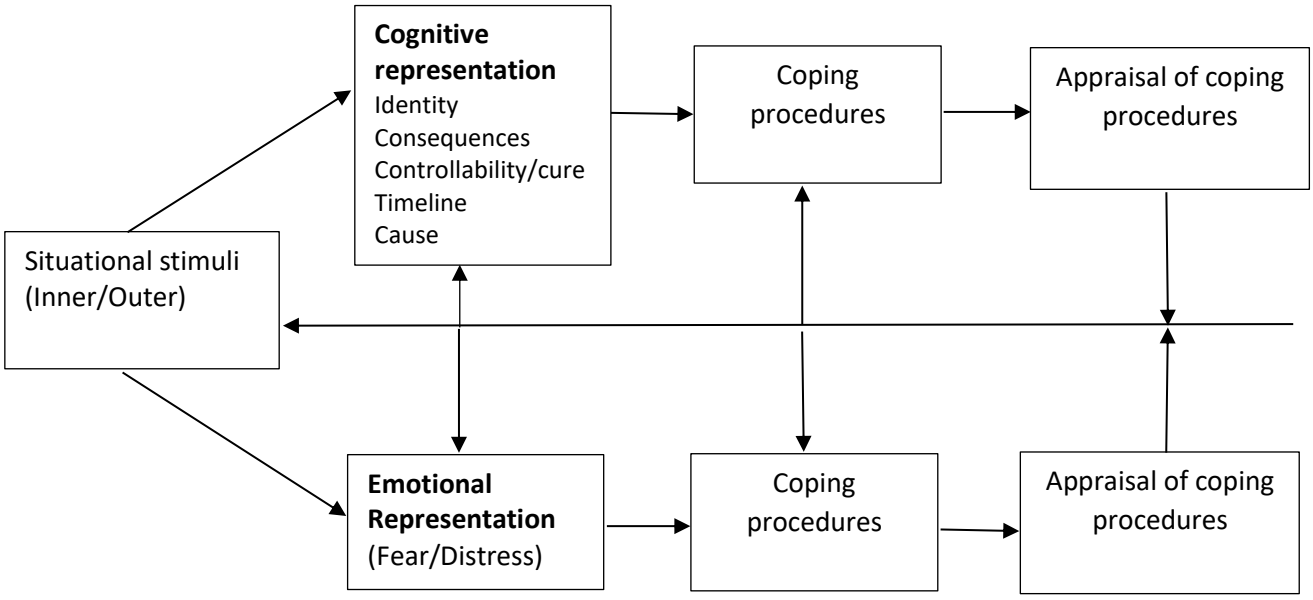
2. The Common-Sense Model of Self-Regulation in Health and Illness

The origin of the Common-Sense Model

The origin of the Common-Sense Model of Self-Regulation in health and illness (CSM) lies in the health communication research that was booming in the 1960s. The goal of this research was to examine if threatening communication about diseases elicited fear that led to attitudes and actions, for preventing the threat [9]. The findings of this research were that fear and the effects of fear were transient. Years later, Leventhal (1970) [10] developed the parallel processing model which posits that processing 'danger control' (i.e., disease threat) occurs parallel and independent from processing 'fear control'. The parallel actions were undertaken and appraised to reduce the negative emotions evoked by health threats (fear control) and reducing the threats themselves (danger control) [11]. In this model a belief about a threat led to an action. Previous research had shown that this belief was not fear because it was known that fear faded away within 24-48 hours. It was the cognitive representation of the disease threat that was the source of motivation for activating plans of action but the content of this cognitive representation was unclear. The first step for the creation of the CSM was undertaken.

Leventhal and colleagues first described the CSM in 1980 [7] and fine-tuned the model afterwards [8,9,12]. The CSM presents how internal stimuli (e.g. symptom experience such as pain) and external stimuli (e.g. disease-related information from family or healthcare professionals) generate cognitive and emotional representations which guide the selection of coping procedures in order to eliminate and control potential or ongoing illness threats. Hereafter, an appraisal of these coping efforts occurs and an evaluation of their success in regulating outcomes (see Figure 1). For example, an individual with a rheumatic disease may view his illness as long lasting but controllable through medication intake, and therefore a change in the medication regimen might be viewed effective to cope with the disease progression. An individual who believes the same disease is uncontrollable and finds it a source of emotional distress may adopt a denial coping response. If the patient appraises a particular coping procedure as being ineffective then this might result in the selection of an alternative coping strategy or even a change in the representation of the illness [9]. So, illness perceptions occur at a two-level stage and are part of a dynamic process.

Figure 1: Common-Sense model of Self-regulation in Health and Illness. Adapted from Leventhal et al. (1997)



Within Leventhal's CSM, cognitive representations are composed out of five main interrelated components that make up patients' views of their illness: 'Timeline' which is the believed time trajectory of the illness; 'Consequences' which includes people's overall evaluation of the seriousness of their conditions, as well as the extent to which these conditions affect specific domains of their lives-physical, social, financial, occupational, etc.; 'Causal attributions' which describes the perceived causal mechanism of the illness; 'Illness Identity': the label a person uses to describe the illness which means

that symptoms are the starting point to label somatic experiences; and 'Control/cure' which measures whether something can be done to control the illness [13]. Researchers found an intercorrelation between the various illness perception dimensions and showed for a number of illnesses that the intercorrelations among the illness perception dimensions were strong and significant, but did not exhibit correlations of a magnitude that was indicative of conceptual overlap [14–16]. Hagger and Orbell (2003) [14] give as a remark in their meta-analysis that the intercorrelations among the dimensions were indicative of a systematic and logical pattern of relations which means that for instance the perception of more severe consequences is associated with a chronic and less controllable disease course.

A closer inspection of the formation of illness perceptions

The first step in establishing and shaping illness perceptions is the interpretation of different sources of information by an individual [15]. Leventhal describes in the CSM three basic sources of information [7,8]. The first source of information is the general pool of 'lay' information already assimilated by the individual from previous social communication and cultural knowledge of the illness. The second source considers information from the external social environment including perceived significant others or authoritative sources, such as family, healthcare professionals, peers. Finally, the individual completes her/his illness representation by taking into account their current experience with the disease. 'Current experience' refers to the somatic or symptomatic information based on current perceptions and previous experiences with the illness. Information from all these sources contributes to the formation of illness representations.

Researchers have looked more in detail at these determinants or correlates of illness perceptions. Following correlates were found as influencing factors of illness perceptions: personality such as optimism and pessimism [15], neuroticism, i.e. negative affectivity, which means experiencing negative emotions such as fear, anger, worry, frustration etc. more likely than average [17], type D personality, which is described as the tendency to experience a high joint occurrence of negative affectivity and social inhibition (i.e., not expressing these negative emotions because of fear of rejection by others) [18,19]; sociodemographic variables such as older age [20–22], female gender [20,22,23], culture [24–26] educational level [20] and high income [22]. Other correlates reported in literature are illness-related variables, the frequency of healthcare visits [15], anxiety, depression or depressive symptomatology [27–29]. Constant and colleagues (2005) [30] mention information from a physician and from the internet as a correlate of illness perceptions.

A clear overview of these correlates or determinants of illness perceptions and the focus on modifiable correlates is lacking. Modifiable correlates are changeable, meaning that they are amenable

for clinical interventions. Knowledge of modifiable correlates is an added value because in some situations it is much more convenient to tackle these correlates instead of the illness perceptions their selves.

Another remark considers that the social environment is less studied in illness perception research, however, not less of importance. Leventhal and colleagues [11] described self-regulation as inherently dependent on the input and expertise of others. Hence, at any specific situation and throughout the individual's development history, every component of the self-regulation system will be shaped and reshaped by the social environment. This means that the contacts throughout the care seeking and treatment process might determine one's experiences with illness [11]. In this regard, it is interesting to know physicians' perceptions concerning the illness of their patients. The fact that illness perceptions of physicians and patients differ or do not differ is not extensively studied in the literature [1,5]. The literature that is available clearly states that healthcare professionals' perceptions are shaped by experience and training and is different from patients' illness perceptions [31]. Furthermore, in clinical practice patients with chronic conditions do not only encounter their treating physicians but also physicians from other specialties who give them likewise advice and information about their condition. Documenting areas of major differences between patients and doctors in their views of an illness can avoid miscommunication and misunderstandings that exist in healthcare [5].

The importance of having knowledge of patients' illness perceptions

Illness perceptions are important because they motivate people to take specific behaviors directed at managing the condition and improve the outcome of their illness [8]. Studies in different patient populations state that illness perceptions are related to important health outcomes such as physical health, recovery, disability, survival/mortality [32], adherence, quality of life, etc. [5,6,14]. Illness perceptions explain, across a range of illnesses, between 25% and 30% of the variance in emotional health outcomes before any coping variables were considered [33]. A growing body of evidence in the past 20 years shows that more 'negative' views of illness held by patients, such as reporting more perceived symptoms, lower perceived controllability of their condition, a longer perceived disease duration and experiencing more serious consequences owing to their conditions, are associated with poorer outcomes, independently of disease-related characteristics or the medical severity of patients' conditions [2,5,6,34]. This research is important because it helps inform clinical interventions and can also provide a basis for identifying patients at an early stage who are at risk of not coping well with their illness [5].

Based on the CSM, researchers assume that illness perceptions generate health outcomes. However, it is possible that these outcomes, in turn, can influence the formation of illness perceptions.

Which means that possibly some outcomes can also be correlates or influencing factors of illness perceptions. For instance, in the case of anxiety: do patients view their illness more negatively because they are anxious, or do they become anxious because they view their illness so seriously and feel they have no control over it? Most of the research conducted in illness perceptions is of cross-sectional nature which does not provide insight into this reciprocal process. The available longitudinal studies did not address this issue until now. Clarity is needed because in designing interventions to alter illness perceptions, clinicians need to know where to intervene [35].

3. Zooming into two severe diseases: systemic sclerosis and systemic lupus erythematosus

In this PhD project we will investigate illness perceptions in patients with complex chronic diseases. As an appropriate example case of patients with complex chronic conditions, we have chosen for a patient population with systemic lupus erythematosus and systemic sclerosis. These two diseases are severe and incurable auto-immune diseases with multiple organ involvement, a heterogeneous occurrence and an unpredictable disease course.

Systemic lupus erythematosus (SLE) is a chronic systemic disease of auto-immune origin with a broad range of clinical manifestations which are diverse and variable, ranging from relatively mild cutaneous and articular involvement through to severe organ damage such as end-stage renal disease and thrombosis [36]. The disease onset is between 18-45 years and it is more prevalent in women than men with a sex ratio of 9:1. The etiology is unknown but probably a combination of hormonal, genetic and environmental factors such as sunlight, drugs, occupational exposure, Epstein-Barr virus, etc. trigger the condition. The prevalence of SLE in Belgium is unknown; the reported values for the incidence and prevalence of SLE vary worldwide. The incidence is 0.3–31.5 cases per 100,000 individuals per year and the prevalence is 3.2–517.5 cases per 100,000 individuals [37]. SLE is more prevalent and even more severe in persons with an African or Asian ancestry, explaining the large prevalence in specific countries or in specific ethnic groups as for instance Africans Americans. SLE patients are at risk for certain comorbidities such as cardiovascular disease or avascular necrosis of bone. Lupus nephritis and central nervous system disease -with possible neuropsychiatric manifestations- might cause serious life threatening complications but also treatment with glucocorticoids and immune suppressant drugs as cyclophosphamide is associated with serious complications [38]. Pregnant women with SLE have a greater risk for complications (miscarriage), disease flares (if active disease at conception) and even congenital lupus with heart block might occur [38]. SLE leads to physical and mental disability [39] characterized by fatigue [40], pain, depression [41], significant cognitive impairment, etc. This disease burden is often associated with poor quality of

life. The treatment options for patients with SLE remain limited compared to those for other rheumatic diseases, such as rheumatoid arthritis [37]. Follow-up is sometimes scattered over many medical specialties dependent of the organs involved. The number of effective treatments for SLE is growing, with the introduction of new (biological) therapies creating new hope [42] .

Systemic sclerosis (SSc) is a rare, auto-immune systemic disease characterized by excessive collagen production (i.e., fibrosis) in the skin and internal organs, vascular damage and inflammation as well autoimmunity [43]. The etiology of SSc is not yet understood although specific environmental triggers such as exposure to silica, solvents, benzene derivates, pesticides, viral triggers, etc. combined with a susceptible genetic background are at the basis of the disease process. Offspring of patients with SSc have a small but definitive risk; at the other part of the spectrum Choctaw American Indians have a high prevalence of SSc [44]. Race is related to a distinct phenotypic profile, with a less favorable outcome for African American patients [43]. It affects more women than men with an average sex ratio of 3:1 [45] and a peak of onset in the fifth decade of life [43]. The prevalence of SSc is <150 per million and the incidence is <10 per million per year in Northern Europe [46]. The mortality rate in SSc is much higher than for other rheumatic diseases [47,48].

Depending on the extent of skin fibrosis/sclerosis, the disease is typically classified in the limited cutaneous subtype (lcSSc) and the diffuse cutaneous subtype (lcSSc). In lcSSc patients, the skin involvement is distal to the elbows and knees with or without face involvement. In dcSSc, the skin thickening is proximal and distal to the elbows and knees with or without facial or truncal involvement [44,49]. Skin fibrosis is a hallmark feature of SSc with worsening skin thickness as a predictor of morbidity and mortality. Patients with dcSSc develop internal organ manifestations during this phase of skin thickening such as renal disease, interstitial fibrotic lung disease, pulmonary arterial hypertension, cardiac disease with cardiac failure and life threatening arrhythmias as well as gastrointestinal involvement (difficulties in swallowing, diarrhea …) and musculoskeletal problems (arthritis, joint rigidity,…) [49]. In addition to the organ impact, patients report also impairments in physical functioning in upper and lower extremities (caused by skin tightness, Raynaud's phenomenon, contractures, etc.) [50–52] and pain (from digital ulcers) [53] which affects their ability to carry out household chores, work and leisure activities. This disease can have specific disfiguring end-stage consequences as amputation of fingers but also specific alterations in the face that might have severe impact on self-esteem.

Treatment of SSc is a challenge because many controversies and uncertainties exist regarding treatment modalities and management lacking evidence based data [47]. Initiatives like the European Scleroderma Trials and Research group (EUSTAR) and Belgian Systemic Sclerosis Cohort (BSSC) try to

stimulate research initiatives in many domains at the European and Belgian level respectively. Patient organizations play an important role in promoting new research initiatives.

Potential hurdles in the care for patients with SLE and SSc in daily practice

In daily practice we are frequently confronted with SLE and SSc patients who report having received contradictory information about their condition or therapy. For instance in the case of pregnancy in SLE, the eventual consequences, risk of miscarriage, intake of drugs like antimalarials to control lupus and glucocorticoids are differently communicated depending of the expert and the organ specialty of the physician. When patients are recently diagnosed with SLE or SSc, most of them received very limited information or have no knowledge of their less prevalent condition. Literature in SSc describes that patients are sometimes dissatisfied because of the limited level of knowledge or understanding of the disease [54].

Not only for the patients who are faced with these devastating conditions, but also for the physicians who care for these conditions, it is not always easy to understand the impact or consequences of such diseases on the patient. When patients are asked about their physicians, they mention uncertainty or diverging advice, often even lack of prescription of an active treatment, and lack of regular follow-up by their treating physicians. This might be experienced by the patient as a lack of interest or even lack of professionality [54].

Because of the complexity and multi-organ involvement of both SLE and SSc, there is in clinical practice often not enough attention for all consequences for the patient and sometimes the focus is limited to a single organ. The uncertainty and lack of knowledge physicians might have can influence the care and management of the patient's condition but also, according to our experience in daily practice, the illness perception of the patient. In our view, sometimes not only what is said or done by the treating physician but even more the way it is done - the non-verbal communication or intonation- can influence the perceptions of the patient.

4. What is already known about illness perceptions in SLE and SSc and their relationship with health outcomes?

Assessment of illness perceptions

Before 1996, illness representations were mainly assessed by means of open ended interviews, and patients' responses were coded into categories [55]. After the introduction of the Illness Perception Questionnaire (IPQ) in 1996 [16], illness perception research boomed. For the first time, the five dimensions of the CSM could be measured in a quantitative way and in large cohorts of patients [56]. Moss-Morris and colleagues created in 2002 a revised version of the IPQ, i.e., IPQ-R [57], including

emotional representations as an additional component because the CSM was proposed by Leventhal and colleagues (1980) [7] as a 'parallel-processing' model which means that people make simultaneous cognitive and emotional representations of their illness. The IPQ-R was also extended with other subscales such as illness coherence which is an overall comprehension of the illness, cyclical timeline perceptions which describes the recurrent pattern of the disease and the control/cure dimension was divided into personal control and treatment control. In 2006, a brief version of the IPQ-R, the brief-IPQ [58] was constructed featuring a single-item scale approach as a response to the long version that was developed before.

Illness perceptions and outcomes in SLE and SSc

A closer inspection of illness perceptions in SLE, shows that patients who perceived more severe consequences, an unpredictable disease course and less comprehension of their disease, reported high levels of depression [59]. The consequences subscale contributed to the greatest degree of variance in depression levels. This has been confirmed by the longitudinal study of Shortall and colleagues (1996) [60], which states that depression, anxiety and self-esteem are predicted by illness perceptions in SLE patients. In the latter study illness perceptions were assessed by a 'problems with SLE' questionnaire. Another SLE study [61] found that illness perceptions are strong predictors of sexual functioning in comparison with medical or socio-demographic characteristics. More specifically, a higher emotional impact of SLE predicted sexual functioning; illness coherence together with emotionality predicted best the subscale 'attractiveness' and personal control predicted best the subscale 'body esteem'. Daleboudt and co-workers [62] found that strong emotional representations were associated with lower self-reported adherence levels. No other associations were found between the other illness perception dimensions and adherence measures. In a recent study of patients with discoid lupus erythematosus [63], which is the cutaneous form of lupus, patients with higher perceived personal control reported better quality of life, lower levels of depression, and better scores of disease activity and severity scores. More concern and emotionality about discoid lupus was associated with worse quality of life, higher depression scores and lower disease activity and severity scores. Kotsis and co-workers (2014) [64] found that the perception of more severe consequences and experiencing more SLE related symptoms were associated with decreased physical functioning.

Concordant findings were found in studies describing illness perceptions in SSc. The most important contributor to good physical health was reporting less severe consequences and a lower score on illness identity, i.e. less SSc related symptoms. Reporting more SSc related symptoms and reacting in a more emotional way to SSc was correlated with worse mental health. Disease related characteristics such as disease activity, disease severity or disease duration explained less variance in physical and mental health in comparison with illness perceptions [65]. Richards et al. (2003) [66]

found that the perception of serious consequences by SSc patients was associated with pain and decreased physical functioning.

5. The objectives and outline of the PhD dissertation

Knowledge regarding illness perceptions is booming. There is an increasing evidence that illness perceptions are important within different aspects of the care and treatment of patients with multisystem diseases. However, there are still many gaps in the literature regarding the concept of illness perceptions, their correlates and impact on outcomes. Therefore, an overall objective of this PhD project was to explore the modifiable correlates of illness perceptions, physicians' perceptions of SLE and SSc and the impact of illness perceptions on health outcomes in patients with SLE and SSc.
To address this overall aim, the thesis is guided by four research questions:

1. What are the modifiable correlates of illness perceptions in patients with chronic somatic conditions? **(Chapter 2)**
2. What are the perceptions of healthcare professionals regarding patients with multisystem diseases?
    a. What are the psychometric properties of a modified illness perception questionnaire for healthcare professionals (IPQ-R HP)? **(Chapter 3)**
    b. What are the perceptions among physicians from the same and other medical disciplines about patients with SLE and SSc? **(Chapter 4)**
3. What are the illness perceptions of patients with SLE and SSc, their rheumatologists and their GPs? **(Chapter 5)**
4. What is the directionality of the associations between illness perceptions and health outcomes in patients with SLE and SSc? **(Chapter 6)**

In Chapter 2, we described the modifiable correlates of illness perceptions in adults with chronic somatic diseases. A systematic review was conducted in order to have an overview of these correlates. Moreover, from the correlates that were found in this study, three were selected for use in Chapter 6. The selected modifiable correlates were also outcomes of illness perceptions.

In Chapter 3, we evaluated the preliminary validation and reliability of an illness perception questionnaire for healthcare professionals, the IPQ-R HP. This instrument was needed to assess perceptions of physicians in the studies described in Chapter 4 and Chapter 5.

In Chapter 4, we analyzed perceptions among physicians from the same and other medical disciplines using vignettes of SLE and SSc patients. This was a multicenter study in physicians from nine different medical disciplines working at three hospitals.

In Chapter 5, we investigated differences and commonalities in illness representations of patients with SLE and SSc, their treating rheumatologists and their general practitioners.

In Chapter 6, we conducted a prospective observational cohort study spanning one year in order to determine the directionality of the associations between illness perceptions and health outcomes in patients with SLE and SSc.

In Chapter 7, we discussed the added value of this thesis, methodological considerations, implications for clinical practice, healthcare policy recommendations and horizons for further research.

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2

MODIFIABLE CORRELATES OF ILLNESS PERCEPTIONS IN ADULTS WITH CHRONIC SOMATIC CONDITIONS: A SYSTEMATIC REVIEW

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Abstract

Objective: When individuals become ill, they want to understand and give meaning to their illness. The interpretation of this illness experience or illness perception is associated with a range of individual, contextual and cultural factors. These correlates of illness perceptions can be categorized in modifiable and not modifiable factors. The purpose of this study was to investigate which modifiable factors were correlated with illness perceptions in adults with chronic somatic diseases.

Methods: All potentially eligible studies identified in four electronic databases were reviewed by 2 independent evaluators, and each relevant article was assessed for methodological quality. Data were extracted, organized and recorded using PRISMA flow diagram. Results were standardized by calculating correlation coefficients.

Results: Fifteen papers met the inclusion criteria. These papers reported a diversity of chronic somatic diseases. We identified 5 groups of modifiable correlates of illness perceptions: illness-related factors, psychosocial factors, medication beliefs, information provision and satisfaction with information and quality of care.

Conclusions: Our findings provide an added value to the knowledge of modifiable factors correlating with illness perceptions such as the importance of psychosocial factors like depression and anxiety and illness-related factors. Knowledge of these correlates can facilitate understanding of patients' illness perceptions and is useful in developing interventions to alter maladaptive illness perceptions.

1. Introduction

Individuals facing illness develop 'common-sense' constructions to conceptualize and give meaning to their illness and its consequences. The search for a meaning is a dynamic process with possible shifts in patients' perceptions and ideas about their illness over time [1,2]. Illness perceptions are beliefs and expectations regarding one's illness that directly influence the individual's cognitive and emotional response to the illness and guide coping strategies to manage illness threat and distress such as for instance seeking social support or problem-focused coping [1,3,4]. Even patients with the same medical condition can hold very disparate views of their illnesses.

Effective and efficient methods to alter dysfunctional illness beliefs, particularly at an early stage of a disease, might generate appropriate coping procedures for managing and living with a chronic disease [5]. At this early stage, the type and quality of the available information which patients use, is crucial in order to give meaning to and manage their illness. Interpreting this information is the first step in the self-regulation of illness which is associated with a range of individual, contextual and cultural factors [6,7]. These factors play an important role in establishing and shaping illness representations [8].

These correlates can be seen as modifiable or non-modifiable. In an exploratory phase, we performed a first literature search in several medical and psychological databases that yielded non-modifiable as well as modifiable correlates associated with illness perceptions. Examples of non-modifiable factors that were significantly correlated with illness perceptions are older age [9–11], female gender [9,11,12] culture [13–15] educational level [9] and high income [11]. Other than the latter demographic factors, the following non-modifiable factors were also significantly related with illness perceptions: neuroticism [16], type D personality [17,18], personal illness experience [19] and symptom reporting [20,21]. Also modifiable correlates of illness perceptions were found such as illness related factors [9], coping style [22], anxiety [20], depression [23] and depressive symptomatology [24], information from a physician and from the internet [22], social functioning and perceived competence [9,25].

So, there is a diversity of literature available about correlates of illness perceptions. The correlates that are non-modifiable such as culture, gender or personality cannot be tackled with clinical interventions, but the illness perceptions that are generated based on these correlates, can. For nurse researchers and clinicians, it is important to have in-depth insight in modifiable correlates, because these can be potential targets for nursing interventions and in that way anticipate the formation of maladaptive illness perceptions. To date, an overview of these modifiable correlates is not available and might therefore be useful for research and clinical purposes. Hence, the purpose of this systematic

review was to investigate which modifiable correlates are associated with illness perceptions in adults with chronic somatic diseases.

2. Methodology

A review protocol based on the PRISMA guidelines [26] for data reporting was developed and was registered in PROSPERO (International prospective register of systematic reviews) with registration number CRD42016047724. Additionally, we complied with the MOOSE guidelines [27].

Literature search and study selection

Medline (via Pubmed), Ebsco host CINAHL, Embase (via embase.com) and Web of Science were searched for eligible articles published between January 1980 and August 2016. A search string for Pubmed was developed and verified by a health sciences librarian. Afterwards, that search string was translated under supervision of the health sciences librarian for use in the other databases. These search strings consisted of text words and controlled vocabulary representing illness perceptions and modifiable correlates (See Appendix 1). For the papers that were eligible for inclusion in this review, backward citations (reference lists) were manually checked for additional relevant articles. The titles and abstracts of citations were independently screened by two reviewers (SA and DDC) and checked for inclusion. Hereafter, full-text papers were screened for eligibility by the latter 2 reviewers and disagreements were resolved by consensus. A third reviewer (RW) was consulted in the case of disagreement.

Domains of illness perceptions

We used 5 core dimensions of illness perceptions as described in Leventhal's Common-Sense Model of health and illness: identity (beliefs concerning illness symptoms; label of the illness), timeline (expected duration), consequences (expected effects on physical, social and psychosocial well-being), cause (causal attributions) and control/cure (to what extent the illness could be controlled or cured) (Leventhal, Nerenz, & Steele, 1984). Subsequent research [29] highlighted the importance of the dimension illness coherence and emotional representations. Illness coherence is the belief that the illness makes sense and was added as a sixth core dimension. Emotional representations, which are the emotional responses of patients generated by the illness and develop in parallel with cognitive representations, were also added.

The most frequently used generic questionnaires that are based on Leventhal's Common-Sense Model, and which are validated and translated into several languages are: the Illness Perception Questionnaire

(IPQ) [30], the revised Illness Perception Questionnaire (IPQ-R) [29] and the Brief Illness Perception Questionnaire (Brief-IPQ) [31].

Inclusion and exclusion criteria

This review included studies that met following inclusion criteria: 1) reporting the modifiable correlates of illness perceptions; 2) using the IPQ, IPQ-R or Brief-IPQ for measuring illness perceptions; 3) using quantitative research designs; 4) including adults with a chronic somatic disease aged 18 years or older; 5) written in English, Dutch, French or German. Exclusion criteria were: 1) psychiatric pathology, dementia, fibromyalgia and chronic fatigue syndrome, as well as studies investigating patient groups with multiple disorders or healthy adults; 2) studies with a qualitative or mixed-methods research design; and 3) editorials, comments, reviews, case reports and letters to the editor and abstracts not accompanied by a full text.

Assessment of methodological quality

The quality of each included studies was assessed using a modified SIGN methodology checklist for cohort studies [32]. We used an adapted version of this checklist where we excluded items that were not applicable for our studies. Concretely, this meant that we selected 6 items (see Supplementary Table 1) which were relevant for observational studies. This checklist was extended with an item about the reporting of potential conflicts of interest. Using this checklist, data concerning methodological quality were independently extracted by two reviewers (SA and DDC) and checked for accuracy. Review authors resolved disagreements through discussion until consensus was reached.

Data extraction and synthesis

Firstly, we scanned the results section for studies that used the IPQ, IPQ-R and Brief-IPQ for operationalizing illness perceptions. Secondly, we looked which modifiable correlates of illness perceptions were reported. We defined modifiable correlates, as correlates that can be changed by clinical interventions. Finally, we reported the results on the correlates we found. The process by which the correlates were grouped into 5 categories was based on thematic analysis [33]. This means that patterns across data were identified, analyzed and reported by two authors until agreement was reached. Also data were gathered from the included studies on sample size, condition studied, location, basic demographics of the population, type of questionnaire for measuring illness perceptions and study design. The correlation coefficients were withdrawn from the studies. If no r-values were reported, results were standardized by converting reported coefficients into r-values [34].

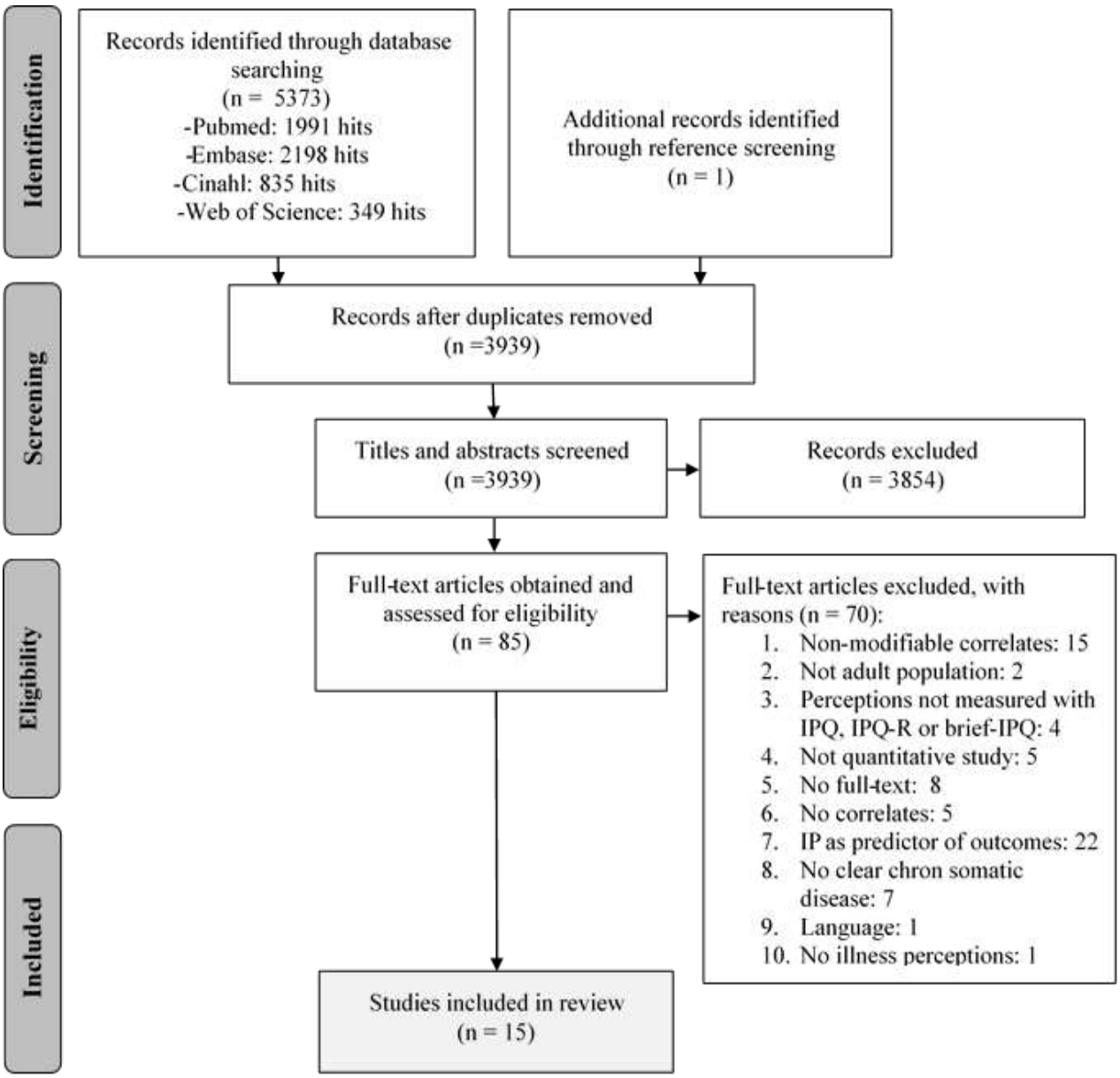
However, for some studies insufficient data were reported to enable the calculation of these effect sizes.

3. Results

Search results

Electronic database searches yielded 5373 articles (Figure 1). After removing duplicates, 3939 articles remained. Of these papers, titles and abstracts were screened on in- and exclusion criteria yielding 85 papers that were subject for full text review. Based on full text review, 70 studies were excluded for different reasons (Figure 1). This process yielded 15 papers to be included in our review.

Figure 1: Flow chart of study selection process



Characteristics of the included studies

An overview of the study characteristics of all included studies can be found in Table 1.

The 15 selected articles encompassed patients with following chronic somatic diseases: chronic obstructive pulmonary disease (COPD) (n= 2) [35,36], psoriasis (n= 2) [37,38], cancer survivors (n= 2) [39,40] coronary heart disease (CHD) (n= 1) [9], acromegaly (n= 1) [41], chronic low back pain (n= 1) [42], sarcoidosis (n= 1) [43], breast cancer (n= 1) [44], chronic cardiovascular disease (n= 1) [45] , rheumatoid arthritis (n= 1) [23], systemic sclerosis (n= 1) [46] and diabetes (n= 1) [47]. A total of 8,619 participants (sample size varied between 49 and 3130) were included. The majority of the studies were cross-sectional (13 studies) and the remaining 2 studies [9,45] had a longitudinal design. Eleven studies took place in Europe, two in Asia, one in US and one in New Zealand. Seven studies used the IPQ-R for measuring illness perceptions, five studies used the Brief-IPQ and three used the IPQ.

Table 1: Study characteristics

Study nr: authors (year)	**Sample size**	**Disease**	**Country**	**Basic demographics**	**Instrument**	**Study design**	**Quality**
Study 1: Aalto, Heijmans, Weinman, and Aro (2005)	N=3130	Coronary heart disease	Finland	50.1% men; mean age 64.4 years (SD=7.0); 9.7 years of education (SD= 2.1)	IPQ	Longitudinal observational	Low risk of bias
Study 2: Andela, Biermasz, Kaptein, Pereira, and Tiemensma (2015)	N=73	Acromegaly	The Netherlands	55% men; mean age 60.1 years (SD=11.6); low education: 40.0%; medium education: 23.0%; high education: 37.0%	IPQ-R	Cross-sectional observational	Low risk of bias
Study 3: Borge, Moum, Lein, Austegard, and Wahl (2014)	N=154	COPD	Norway	51.3% men; mean age 64.6 years (SD=10.2); low education: 40.2%; medium education: 30.5% ; high education: 27.9%	Brief-IPQ	Cross-sectional observational	Low risk of bias
Study 4: Fortune, Richards, Main, and Griffiths (1998)	N=162	Psoriasis	UK	51.9% men; mean age 42 years (SD=14.0); no data on educational level	IPQ	Cross-sectional observational	High risk of bias
Study 5: Heyduck, Meffert, and Glattacker (2014)	N=201	Chronic low back pain	Germany	35.8% men; mean age 54.1 years (SD=11.4); elementary school: 42.8%; secondary school: 16.4%; technical school: 23.9%; university: 11.9%; no certificate: 2.5%	IPQ-R	Cross-sectional observational	Low risk of bias
Study 6: Howard, Hallas, and Carby (2009)	N=59	COPD	UK	39% men; mean age 62.4 years (SD=12.0); secondary school: 65.0%	IPQ-R	Cross-sectional observational	Low risk of bias
Study 7: Husson et al. (2013)	N=3080	Cancer survivors	The Netherlands	56.3% men; mean age 62.6 years (SD=11.02); primary school: 23.9% ; secondary school: 32.6%; intermediate school: 28.8%; university: 14.6%	Brief-IPQ	Cross-sectional observational	Low risk of bias
Study 8: Ireland & Wilsher (2010)	N=81	Sarcoidosis	New-Zealand	48% men; mean age: 49 years (SD=13.0); no data on educational level	IPQ-R	Cross-sectional observational	Low risk of bias

Study 9: Iskandarsyah et al. (2013)	N=70	Breast cancer	Indonesia	100% women; mean age: 45.6 years (SD=7.9); no education: 10%; elementary: 51%; junior high school: 16%; senior high school: 14%; college/university: 9%	Brief-IPQ	Cross-sectional observational	Low risk of bias
Study 10: Karademas, Paschali, Hadjulis and Papadimitriou (2016)	N=119	Chronic cardiovascular disease	Greece	69.9% men; mean age: 62.4 years (SD=12.9); no data on educational level	IPQ-R	Longitudinal observational	Low risk of bias
Study 11: Murphy, Dickens, Creed and Bernstein (1999)	N=62	Rheumatoïd arthritis	UK	16.1% men; median age: 59.5 years (IQR=50.8–68); no data on educational level	IPQ	Cross-sectional observational	Low risk of bias
Study 12: Richards et al. (2003)	N=49	Systemic Sclerosis	UK	14% men; mean age: 53 years (SD=12.0); no data on educational level	IPQ-R	Cross-sectional observational	Low risk of bias
Study 13: Thomas et al. (2014)	N=89	Diabetes	USA	39.3% men; age 65 or older: 50.5 %; no degree: 32.5%; high school diploma: 28.1%; associate's degree: 4.5%; bachelor's degree: 16.8%; graduate degree: 18.1%	Brief-IPQ	Cross-sectional observational	Low risk of bias
Study 14: Wahl et al. (2014)	N=254	Psoriasis	Norway	60% men; mean age: 47 years (SD=12.0); primary school: 60%; university < 4 years: 20%; university ≥ 4 years: 19%	IPQ-R	Cross-sectional observational	Low risk of bias
Study 15: Zhang et al. (2016)	N= 1036	Cancer survivors	Hong Kong	40.1% men; mean age: 55.2 years (SD=11.9); no formal educational level: 5.1%; primary educational level: 28.3%; secondary educational level: 52.4%; tertiary educational level: 14%; missing: 0.2%	Brief-IPQ	Cross-sectional observational	Low risk of bias

COPD: chronic obstructive pulmonary disease; IPQ: illness perception questionnaire; IPQ-R: revised illness perception questionnaire; Brief-IPQ: brief illness perception questionnaire

Methodological quality

Thirteen of the fifteen studies had an appropriate focused clinical question. The selection of the cohorts from comparable source populations was clearly described in 13 of the 15 articles and the participation rate was reported in 14 of 15 studies. In all studies, the outcomes were clearly defined using reliable and valid methods. Confidence intervals were reported in 14 of 15 studies. The main potential confounders were identified and taken into account in the design and analyses of all except for three studies. Four of the 15 studies did not report potential conflicts of interest (see Supplementary Table 1).

Modifiable correlates of illness perceptions

The included studies identified 5 broad groups of correlates of illness perceptions: medication beliefs, information provision & satisfaction, quality of care, illness-related factors and psychosocial factors. An overview of these modifiable factors and its correlation with illness perception dimensions is provided in Tables 2-6. The correlations with the causal attributions are not shown in the tables.

Medication beliefs (see Table 2)

Regarding medication beliefs, three modifiable correlates were identified. In patients with acromegaly, general harm [41], i.e. the beliefs that medicines are poisonous, addictive or harmful and general overuse [41], i.e. the beliefs that medicines are overprescribed, were correlated with the emotional representation dimension which means that the greater the belief that there is general harm ($r= 0.28$, $p<0.05$) or general overuse of the medication ($r= 0.27$, $p<0.05$), the higher the emotional impact on the patient. A third factor, being the necessity of taking the medication from a medical perspective, which was studied in patients with sarcoidosis [43] was negatively correlated with emotional representations ($r= -0.31$, $p<0.05$), the perceived consequences of the disease ($r= -0.25$, $p<0.05$) and perceiving the time course as stable and less recurrent ($r= -0.38$, $p<0.01$). Higher scores on general harm were also associated with higher psychological attributions ($r= 0.24$, $p<0.05$) and higher perceived risk factors ($r= 0.24$, $p< 0.05$) as a cause of the disease (data not shown in table).

Information provision and satisfaction (see Table 3)

Cancer survivors receiving information about the disease [39], perceived a higher personal control ($r= 0.11$, $p<0.01$). Receiving information about care services in these patients [39] was correlated with more symptoms characterized with the disease ($r= 0.13$, $p<0.01$), more severe consequences ($r= 0.13$, $p<0.01$),

controllability by treatment (r= 0.06, p<0.01), concern (r= 0.08, p<0.01) and emotionality (r= 0.10, p<0.01). In cancer survivors, a lower satisfaction with received information [39] was correlated with more symptoms characterized with the disease (r= -0.16, p<0.01), perception of more severe consequences (r= -0.23, p<0.01), more controllability by treatment (r= -0.11, p<0.01), perception of a chronic time course (r= -0.10, p<0.01), more understanding (r= -0.10, p<0.01), more concern (r= -0.19, p<0.01) and more emotional representations (r= -0.24, p<0.01). In breast cancer patients, satisfaction with the amount and content of information [44] was related with less emotionality (r= -0.25, p<0.05). Also in this patient population, satisfaction with form and timing of information [44] was correlated with less controllability of breast cancer by the patient herself (r= -0.24, p<0.05), less understanding of the disease (r= -0.25, p<0.05), less concern (r= -0.36, p<0.01) and less emotional impact (r= -0.33, p<0.01). Knowledge about psoriasis [38] was correlated with the perception that psoriasis is chronic (r= 0.24, p<0.001), understandable (r= 0.27, p<0.001) and having less emotional impact on the patient (r= -0.17, p<0.01).

Quality of care (see Table 4)

Diabetes patients, reporting higher perceived chronic care quality also indicated better disease understanding (r= 0.24, p<0.05) [47]. In chronic back pain patients, trust in the physician [42] was related with perceiving less severe consequences to the disease (r= -0.18, NS).

Illness-related factors (see Table 5)

In COPD patients, breathlessness [35] was related with more symptoms characterized with the disease (r= 0.62, p<0.001), severe consequences (r= 0.67, p<0.001) and more concern (r= 0.44, p<0.01) and emotionality (r= 0.45, p<0.001). In lung testing, FEV1% [35,36]**,** -i.e. the percent predicted forced expiratory volume in the first second- was correlated with ~~a~~ less symptoms characterized with the disease (r= -0.16, NS; r= -0.48, p<0.001), less perceived consequences (r= -0.32, p<0.05; r= -0.43, p<0.001), less chronic time course (r= -0.19, p<0.05), less concern (r= -0.24, p<0.001) and a low emotional impact of the disease (r= -0.21, p<0.01). In psoriasis patients, the PASI (Psoriasis Area and Severity Index) was correlated with experiencing more severe consequences due to the disease (r= 0.21, p<0.01) [38], less personal control (r= -0.39, p<0.05) [37], a chronic time course (r= 0.17, p<0.001) (Wahl et al., 2014) and emotionality (r= 0.13, p<0.05) [38]. Physical symptom distress [40] -which is any distress associated with a particular symptom- was correlated with all dimensions except for controllability by treatment. Pain in systemic sclerosis patients [46] was related with more symptoms characterized with the disease (r= 0.36, p<0.05), high personal control (r= 0.54, p<0.01) and high emotionality (r= 0.38, p<0.05). CHD risk factors

and CHD related comorbidities [9] were correlated with more symptoms characterized with the disease (r= 0.06, p<0.01; r= 0.19, p<0.001, respectively), perceived severe consequences (r= 0.08, p<0.001; r= 0.14, p<0.001, respectively) and a chronic time course (r= 0.09, p<0.001; r= 0.09, p<0.001, respectively). CHD related comorbidities were only correlated with experiencing less personal control (r= -0.11, p<0.001).

Concerning the correlations with causal attributions, CHD risk factors were associated with epidemiological risk factors (r= 0.22, p<0.001) and CHD comorbidities with stress (r= 0.08, p<0.001) and external factors (i.e. poor medical care, germ/virus, pollution, other people, chance/luck and diabetes) (r= 0.06, p<0.001), health behavior (r= 0.06, p<0.01) and epidemiological risk factors (r= 0.07, p<0.001).

Psychosocial factors (see Table 6)

In patients with CHD, higher perceived competence [9] -i.e. confidence in one's personal competence in successfully attaining important life goals- was associated with less symptoms characterized with the disease (r= -0.18, p<0.001), less severe consequences (r= -0.31, p<0.001), high personal control (r= 0.21, p<0.001) and a chronic disease course (r= 0.08, p<0.001). Receiving social support in CHD patients [9] was associated with less symptoms characterized with the disease (r= -0.10, p<0.001) and a higher personal control (r= 0.11, p<0.001).

Depression in rheumatoid arthritis and COPD was associated with more symptoms characterized with the disease (r= 0.31, p<0.05) (Murphy et al., 1999), less personal control (r= -0.40, p<0.01) (Murphy et al., 1999), having an emotional impact on the patient (r= 0.53, p<0.001) [36], and perceiving severe consequences (r= 0.47, p<0.001; r= 0.48, p<0.001) [23,36], which was also reported in sarcoidosis patients (r= 0.47, p<0.001) [43]. In sarcoidosis patients [43], anxiety was associated with having a clear picture of the disease (r= 0.29, p<0.05). In COPD patients [36], anxiety was related with perceiving severe consequences (r= 0.43, p<0.001) and having an emotional impact (r= 0.61, p<0.001). Also in the latter patient population, panic severity was associated with perceiving severe consequences (r= 0.77, p<0.001) and having a high emotional impact (r= 0.75; p<0.001). Maladaptive health beliefs (Karademas et al., 2016) were negatively correlated with personal control (r= -0.25, p<0.01) and illness coherence (r= -0.22, p<0.05) and positively correlated with the biological causes a patient attributes to his/her illness to (r= 0.31, p<0.01).

Concerning the causal attributions: perceived competence and social support in CHD patients [9] were associated with stress (r= -0.21, p<0.001; r= -0.13, p<0.001, respectively), external factors (i.e. poor medical care, germ/virus, pollution, other people, chance/luck and diabetes) (r= -0.18, p<0.001; r= -0.08, p<0.001, respectively). Only perceived competence was also associated with life-course (r= -0.24, p<0.001)

epidemiologic factors (r= -0.10, p<0.001), health behavior (r= -0.11, p<0.001) and internal factors such as the patients' own behavior, mental attitude and personality (r= -0.21, p<0.001).

Table 2: Medication beliefs

Modifiable correlate	Identity	Consequences	Personal control	Treatment control	Timeline acute/chronic	Timeline cyclical	Illness coherence	Concern	Emotional Representations
General harm	r= 0.05[2]	r= 0.01[2]	r= 0.14[2]	r= -0.11[2]	r= -0.19[2]	r= -0.02[2]	r= 0.01[2]	/	r= 0.28*[2]
General overuse	r= -0.04[2]	r= -0.04[2]	r= 0.14[2]	r= -0.11[2]	r= -0.20[2]	r= 0.07[2]	r= -0.01[2]	/	r= 0.27*[2]
Necessity for medication	/	r= -0.25*[8]	/	/	/	r= -0.38†[8]	/	/	r= -0.31*[8]

*: p<0.05; †: p<0.01; numbers in superscript refer to study number

Table 3: Information provision and satisfaction

Modifiable correlate	Identity	Consequences	Personal control	Treatment control	Timeline acute/chronic	Timeline cyclical	Illness coherence	Concern	Emotional Representations
Info about disease	r= 0.05[7]	r= 0.02[7]	r= 0.11†[7]	r= -0.10[7]	r= -0.04[7]	/	r= -0.12[7]	r= -0.05[7]	r= 0.03[7]
Info about medical test	r= -0.02[7]	r= 0.01[7]	r= -0.01[7]	r= -0.05[7]	r= 0.03[7]	/	r= -0.03[7]	r= 0.04[7]	r= -0.04[7]
Info about treatment	r= -0.04[7]	r= 0.02[7]	r= -0.01[7]	r= -0.03[7]	r= -0.01[7]	/	r= -0.04[7]	r= 0.04[7]	r= 0.06[7]
Info about care services	r= 0.13†[7]	r= 0.13†[7]	r= -0.04[7]	r= 0.06*[7]	r= 0.04[7]	/	r= 0.05[7]	r= 0.08†[7]	r= 0.10†[7]
Satisfaction with information	r= -0.16†[7]	r= -0.23†[7]	r= -0.05[7]	r= -0.11†[7]	r= -0.10†[7]	/	r= -0.10†[7]	r= -0.12†[7]	r= -0.24†[7]
Satisfaction with amount & content info	r= -0.08[9]	r=-0.08[9]	r= -0.01[9]	r=-0.08[9]	r= -0.12[9]	/	r= -0.09[9]	r= -0.09[9]	r= -0.25*[9]
Satisfaction with form & timing info	r= -0.10[9]	r= -0.05[9]	r= -0.24*[9]	r= -0.21[9]	r= -0.02[9]	/	r= -0.25*[9]	r=-0.36†[9]	r= -0.33†[9]
Knowledge about disease	/	Insufficient data[14]	/	/	r= 0.24‡[14]	/	r= 0.27‡[14]	/	r= -0.17†[14]

*: p<0.05; †: p<0.01; ‡: p<0.001; numbers in superscript refer to study number

Table 4: Quality of care

Modifiable correlate	Identity	Consequences	Personal control	Treatment control	Timeline acute/chronic	Timeline cyclical	Illness coherence	Concern	Emotional Representations
Perceived chronic care quality	r= -0.13[13]	r= -0.14[13]	r= 0.10[13]	r=0.22[13]	r= -0.03[13]	/	r= 0.24*[13]	r= -0.06[13]	r= -0.12[13]
Trust in physician	/	r= -0.18[5]	/	/	/	/	/	/	/

*: p<0.05; numbers in superscript refer to study number

Table 5: Illness-related factors

Modifiable correlate	Identity	Consequences	Personal control	Treatment control	Timeline acute/chronic	Timeline cyclical	Illness coherence	Concern	Emotional Representations
Breathlessness	r= 0.62‡[3]	r= 0.67‡[3]	/	/	/	/	/	r= 0.44†[3]	r=0.45‡[3]
FEV1%	r= -0.16[6] r= -0.48‡[3]	r= -0.32*[6] r= -0.43‡[3]	r= -0.06[6] /	r= -0.08[6] /	r= -0.23[6] r= -0.19*[3]	r= -0.01[6] /	r= -0.10[6] /	 r= -0.24‡[3]	r=-0.07[6] r= -0.21†[3]
PASI	/	r= 0.21†[14]	r= -0.39*[14]	/	r= 0.17‡[14]	/	No data[14]	/	r= 0.13*[14]
Physical sympt distress	r= 0.32†[15]	r= 0.40 †[15]	r= -0.08*[15]	r= -0.06[15]	r= 0.40†[15]	/	r= -0.08†[15]	r= 0.29†[15]	r= 0.39†[15]
Pain	r= 0.36*[12]	r= 0.54†[12]	r= -0.06[12]	r= -0.11[12]	r= 0.22[12]	r= 0.19[12]	r= -0.25[12]	/	r= 0.38*[12]
CHD risk factors	r= 0.06†[1]	r= 0.08‡[1]	r= 0.01[1]	/	r= 0.09‡[1]	/	/	/	/
CHD comorbidities	r= 0.19‡[1]	r= 0.14‡[1]	r= -0.11‡[1]	/	r= 0.09‡[1]	/	/	/	/

PASI: Psoriasis Area and Severity Index; FEV1%: percent predicted forced expiratory volume in the first second; CHD: coronary heart disease; *: p<0.05; †: p<0.01; ‡: p<0.001; numbers in superscript refer to study number

Table 6: Psychosocial factors

Modifiable correlate	Identity	Consequences	Personal Control	Treatment control	Timeline acute/chronic	Timeline cyclical	Illness coherence	Concern	Emotional Representations
Perceived competence	r= -0.18‡[1]	r= -0.31‡[1]	r= 0.21‡[1]	/	r= 0.08‡[1]	/	/	/	/
Social support	r= -0.10‡[1]	r= -0.05[1]	r= 0.11‡[1]	/	r= -0.01[1]	/	/	/	/
Depression	r= 0.31*[11] r= 0.20[6] /	r= 0.48‡[11] r= 0.47‡[6] r= 0.47‡[8]	r= -0.40†[11] r= -0.18[6] /	/ r= -0.02[6] /	r= 0.06[11] r= 0.21[6] /	/ r=0.11[6] /	/ r= -0.01[6] /	/ /	/ r= 0.53‡[6] /
Anxiety	/ r=0.23[6]	/ r=0.43‡[6]	/ r=0.09[6]	/ r=0.09[6]	/ r=0.10[6]	/ r=0.24[6]	r= 0.29*[8] r=0.02[6]	/	/ r=0.61‡[6]
Maladaptive health beliefs	/	r=0.12[10]	r= -0.25†[10]	r= -0.25†[10]	r=0.04[10]	r=0.14[10]	r= -0.22*[10]	/	r= 0.02[10]
Panic severity	r= 0.41[6]	r=0.77‡[6]	r= 0.12[6]	r=-0.10[6]	r=0.18[6]	r=0.40[6]	r=0.07[6]	/	r= 0.75‡[6]

*: p<0.05; †: p<0.01; ‡: p<0.001; numbers in superscript refer to study number

4. Discussion

This systematic review identified 5 categories of modifiable correlates of illness perceptions: medication beliefs; information provision and satisfaction; quality of care; illness-related factors and psychosocial factors.

We found that medication beliefs were correlated with emotional representations but not with cognitive beliefs. Patients' representations of illness and treatment show a 'common-sense' logic, even when treatment evaluations are based on misconceptions they appear to draw on [48]. Concerns about prescribed medication are not only related to side effects but also to more general concerns, even when medication is well tolerated. These concerns are often related to beliefs about the negative effects of medication such as long-term health effects, drug dependence, medication cost and dislike of having to rely on medicines [49]. Horne & Weinman (2002) [49] suggest that one strategy to shift patients' beliefs about the necessity of their treatment is through their illness perceptions. Unfortunately, most studies investigating the relationship between medication beliefs and illness perceptions are cross-sectional. Hence, the directionality of the associations is not clear.

Hagger & Orbell (2003) [6] describe that forming illness perceptions is influenced by somatic and symptomatic information and the information that patients receive from healthcare professionals. In the present study, we found that not only the information a patient gets, but also the satisfaction with the information received, is a correlate of illness perceptions. Satisfaction with information has been shown to influence long-term outcomes after treatment such as depression and mental component scores of reported health [50]. Research has suggested that it is the inferences individuals make about the information that determine levels of distress rather than the meanings the information giver intends to convey. Hence, people interpret the information they have been given within their own framework of ideas and theories of their illness [51]. So, it is likely to first access patients' views about their illness and treatments in relation to their satisfaction with information prior to and during their treatment.

A closer look at the psychosocial factors shows that they are together with illness-related factors the most influencing factors which need to be tackled first in designing interventions. The correlation of depression and anxiety with illness perceptions was reported in four studies describing psychosocial factors. A recent meta-analysis [52] showed that perceptions of illness consequences and emotional representations had the strongest relationship with depression, anxiety and quality of life as seen in our study. This strong relationship between emotional representations and outcomes such as anxiety and depression raises the question about measuring comparable concepts. The differences between these two concepts are that emotional representations are specific to the illness under investigation whereas anxiety

and depression are measured more generically. However, their interdependence poses a problem in interpreting correlations.

This review has some strengths and limitations. A strength of this review was that all the included articles defined illness perceptions by using Leventhal's common-sense model. Hence, it was possible to generalize the results although the correlates were found in different patient populations. However, most studies using one of the aforementioned illness perception questionnaires, studied causal factors poorly probably because of the more time consuming analysis process. Another strength was that the methodological quality of the included studies was rated overall as good. A possible limitation might be that some relevant papers were missed despite our use of a search string per database, carefully developed in collaboration with a health sciences librarian or because there was a predefined selection of instruments for assessing illness perceptions (which was stated as an inclusion criterion). Secondly, no meta-analysis could be conducted due to the low number of studies investigating a specific correlate, the large variability in diseases and the lack of sufficient raw data.

This study gives more insight in the correlates of illness perceptions that are modifiable and that can be handled by healthcare professionals. It supplies information in the form of an outline to guide healthcare professionals in the communication and education of their patients in daily clinical practice. For instance, a checklist with modifiable correlates can be an addition to the Representational Approach to patient education of Donovan and colleagues (2007)[53]. This approach requires eliciting and understanding patients' pre-existing representations of illness before giving new information. In this way, healthcare professionals and patients have the opportunity to recognize gaps, confusions, and misconceptions in the patient's representation [53]. Knowledge about the most influential correlates, i.e. anxiety, depression, illness-related factors, can be added to the different steps in this Representational Approach to patient education and tailored to each patient individually. Moreover, the first and second step that describes the representational assessment and identification or exploration of gaps, errors and confusions can be extended by the correlates found in this study. The modifiable correlates that are relevant for a specific patient situation can be withdrawn and can facilitate the patient education process.

The findings of the current systematic review also show important implications for future research. Firstly, knowledge about the modifiable correlates of illness perceptions also applies to research concerning adjustment to chronic illness. Working models for adjustment to chronic illness such as described by Moss-Morris (2013) [54] can be extended by filling out these correlates and more specifically the most influential correlates such as illness-related factors and psychosocial factors. For example personal background factors can be extended by adding depression and anxiety.

Secondly, more qualitative research is needed. Qualitative research provides an in-depth understanding to get more insight in the factors that are considered by patients themselves as correlates. These factors can be patient specific factors but also factors at the level of the health care provider, or the healthcare system. This design makes it possible to go in detail about why people think that certain correlates contribute to certain illness perceptions. Qualitative research about illness perceptions is available [55] but research about modifiable correlates is scarce. More qualitative research about the correlates of illness perceptions would add to the understanding of illness perceptions.

Thirdly, the results of this review can make an attempt to address theoretical questions based on Leventhal's Self-Regulation model. In this theory, illness perceptions are the building blocks of inquiry and intervention for altering and tackling 'undesired' outcomes. One important issue that can be discussed and investigated is that -in some situations- it is probably much more convenient to tackle the modifiable correlates of illness perceptions for altering outcomes than the illness perceptions per se. Hence, an overview of the types and characteristics of the correlates of illness perceptions is necessary which makes planning an intervention protocol much more easy.

Fourthly, we found that some modifiable correlates are also outcomes of illness perceptions. The correlates depression, anxiety and illness-related factors, medication beliefs can be both determinants and outcomes of illness perceptions raising concern about the directionality of these relationships. Further longitudinal research needs to clarify the directionality of the associations between modifiable correlates and illness perceptions.

We can **conclude** that our findings provide an added value to the knowledge about illness perceptions such as the importance of psychosocial factors like depression and anxiety and illness-related factors. Our findings could facilitate understanding of patients' illness perceptions, enhances patient-provider communication and can be useful in developing effective interventions and tailored patient education programs to alter maladaptive illness perceptions and ultimately to improve care.

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Appendix 1: Search strings of the 4 databases

1. Pubmed

("Perception"[Mesh:NoExp] OR illness perception*[tiab] OR illness cognition*[tiab] OR perceived illness*[tiab] OR leventhal[tiab] OR illness belief*[tiab] OR illness representation*[tiab] OR illness schema[tiab] OR illness model*[tiab] OR "health appraisal"[tiab] OR "health perception*"[tiab] OR "patient perception*"[tiab] OR "self regulation"[tiab] OR "self regulatory"[tiab] OR "common sense model"[tiab]) AND ("Adult"[Mesh] OR adult*[tiab] OR elder*[tiab] OR grown-up*[tiab] OR aged pat*[tiab] OR aged pers*[tiab]OR aged people[tiab] OR aged person[tiab] OR elderly patient[tiab] OR elderly people [tiab]OR elderly person*[tiab]) AND ("Chronic Disease"[Mesh] OR chronic disease*[tiab] OR chronic disorder*[tiab] OR chronic ill*[tiab] OR chronically ill*[tiab] OR persistent ill*[tiab] OR chronic patient* OR persistent disease*[tiab] OR persistent disorder*[tiab] OR systemic disorder*[tiab] OR systemic disease*[tiab] OR "Patient Acuity"[Mesh] OR Patient Acuit*[tiab] OR illness [tiab]) AND (correlat*[tiab] OR "Association"[Mesh:NoExp] OR associat*[tiab] OR determinant*[tiab] OR Influencing factor*[tiab] OR relat*[tiab])

2. Embase

('perception'/de OR 'illness perception*':ab,ti OR 'illness cognition*':ab,ti OR 'perceived illness*':ab,ti OR 'leventhal':ab,ti OR 'illness belief*':ab,ti OR 'illness representation*':ab,ti OR 'illness schema':ab,ti OR 'illness model*':ab,ti OR "health appraisal":ab,ti OR "health perception*":ab,ti OR "patient perception*":ab,ti OR 'self regulation'/de OR "self regulation":ab,ti OR "self regulatory":ab,ti OR "common sense model":ab,ti) AND ('adult'/de OR 'adult':ab,ti OR 'grown-up*':ab,ti OR 'grownup':ab,ti OR 'grownups':ab,ti OR 'aged patient':ab,ti OR 'aged people':ab,ti OR 'aged person':ab,ti OR 'elderly patient':ab,ti OR 'elderly people':ab,ti OR 'elderly person':ab,ti) AND ('chronic disease'/de OR 'chronic disease*':ab,ti OR 'chronic illness':ab,ti OR 'chronic disorder*':ab,ti OR 'chronic patient'/de OR 'chronic patient*':ab,ti OR 'persistent ill*':ab,ti OR 'persistent disease*':ab,ti OR 'persistent disorder*':ab,ti OR 'systemic disease'/de OR 'systemic disease*':ab,ti OR 'patient acuity'/de OR 'patient acuit*':ab,ti OR 'illness':ab,ti) AND ('correlat*':ab,ti OR 'association'/de OR 'associat*':ab,ti OR 'determinant*':ab,ti OR 'influencing factor*':ab,ti OR 'relat*':ab,ti)

3. Web of Science

TS=("perception" OR "illness perception*" OR "illness cognition*" OR "perceived illness*" OR "leventhal" OR "illness belief*" OR "illness representation*" OR "illness schema" OR "illness model*"OR "health appraisal" OR "health perception*" OR "patient perception*" OR "self regulation" OR "self regulatory" OR "common sense model") AND TS=("adult" OR "grown-up*" OR "grownup*" OR "aged patient" OR "aged people" OR "aged person" OR "elderly patient" OR "elderly people" OR "elderly person") AND TS=("chronic disease*" OR "chronic disorder*" OR "chronic illness"OR "chronic patient" OR "persistent ill*" OR "persistent disease*" OR "persistent disorder" OR "systemic disease" OR "Patient Acuit*" OR illness) AND TS=("correlate*" OR "associate*" OR "determinant*" OR "influencing factor*" OR "relat*")

4. Cinahl

((MH "Perception") OR (TI illness perception* OR AB illness perception*) OR (TI illness cognition* OR AB illness cognition*) OR (TI perceived illness* OR AB perceived illness*) OR (TI leventhal OR AB leventhal) OR (TI illness belief* OR AB illness belief*) OR (TI illness representation* OR AB illness representation*) OR (TI illness schema OR AB illness schema) OR (TI illness model* OR AB illness model*) OR (TI "health appraisal" OR AB "health appraisal") OR (TI "health perception*" OR AB "health perception*") OR (TI "patient perception*"OR AB "patient perception*") OR (MH "Self Regulation") OR (TI "self regulation" OR AB "self regulation") OR (TI "Self regulatory" OR AB "Self regulatory") OR (TI "common sense model" OR AB "common sense model")) AND ((MH"Adult") OR (TI adult OR AB adult) OR (TI grownups OR AB grownups) OR (TI grown-up* OR AB grown-up*) OR (TI grownup OR AB grownup)OR (TI aged patient OR AB aged patient) OR (TI aged people OR AB aged people) OR (TI aged person OR AB aged person) OR (TI elderly patient OR AB elderly patient) OR (TI elderly people OR AB elderly people)OR (TI elderly person OR AB elderly person)) AND ((MH "chronic disease") OR (TI chronic disease* OR AB chronic disease*) OR (TI chronic illness OR AB chronic illness) OR (TI chronic disorder* OR AB chronic disorder*) OR (TI chronic patient* OR AB chronic patient*) OR (TI persistent ill* OR AB persistent ill*) OR (TI persistent disease* OR AB persistent disease*) OR (TI persistent disorder* OR AB persistent disorder*) OR (TI systemic disease* OR AB systemic disease*) OR (TI patient acuit* OR patient acuit*) OR (TI illness OR AB illness)) AND ((TI correlat* OR AB correlate*) OR (TI associat* OR AB associate*) OR (TI determinant* OR AB determinant*) OR (TI influencing factor* OR AB influencing factor*) OR (TI relat* OR AB relat*))

Supplementary Table 1: Methodological quality appraisal using the SIGN methodology checklist for cohort studies

	Aalto, Heijmans, Weinman, and Aro (2005)	Andela, Biermasz, Kaptein, Pereira, and Tiemensma (2015)	Borge, Moum, Lein, Austegard, and Wahl (2014)	Fortune, Richards, Main, and Griffiths (1998)	Heyduck, Meffert, and Glattacker (2014)	Howard, Hallas, and Carby (2009)	Husson et al. (2013)	Ireland and Wilsher, 2010	Iskandarsyah et al., 2013	Karademas, Paschali, Hadjulis and Papadimitriou (2016)	Murphy, Dickens, Creed and Bernstein (1999)	Richards et al. (2003)	Thomas et al. (2014)	Wahl et al. (2014)	Zhang et al. (2016)
Appropriate focused clinical question	+	+	+	−	+	+	+	+	+	−	+	+	+	+	+
Cohorts are selected from comparable source populations	+	+	+	+	+	+	+	+	?	+	+	?	+	+	+
Participation rate is reported in each of the groups studied	+	+	+	−	+	+	+	+	+	+	+	+	+	+	+
Clearly defined outcomes measured using reliable and valid methods	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Confidence intervals are reported	+	+	+	−	+	+	+	+	+	+	+	+	+	+	+
The main potential confounders are identified & taken into account in the design and analysis	+	+	+	−	+	+	+	+	+	+	+	−	−	+	+
Potential conflict of interest reported	+	+	+	+	+	−	+	−	+	+	+	−	−	+	+

Symbols indicate: + : low risk of bias; − :high risk of bias; ? : unclear risk of bias

3

DEVELOPMENT AND PRELIMINARY EVALUATION OF THE VALIDITY AND RELIABILITY OF A REVISED ILLNESS PERCEPTION QUESTIONNAIRE FOR HEALTHCARE PROFESSIONALS

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Abstract

Objective: Diverging perceptions between individual patients with somatic diseases and their healthcare professionals might cause problems in communication and decision-making. To date, no measurement tool is available to compare the illness perceptions between these two groups. The Revised Illness Perception Questionnaire (IPQ-R) is a validated, widely used instrument in many patient populations with somatic conditions. The aim of this study was to adapt the IPQ-R to a healthcare professional's version (IPQ-R HP) and to perform a preliminary evaluation of its validity and reliability.

Methods: After adaptation of the IPQ-R HP, 17 doctors from 3 general hospitals and 9 head nurses from a university hospital evaluated the face and content validity of the IPQ-R HP. The results were quantified using the content validity index (CVI) and a modified kappa index (k*). For the reliability measurements a group of nurses from 4 nursing wards participated at 2 time points with an interval of 4 weeks. Internal consistency and test-retest reliability were calculated.

Results: Twenty-eight of the 38 items demonstrated excellent content validity and four items showed good content validity. Four items had a sufficient k* and two items had a low CVI. The average CVI of the 7 dimensions ranged from 0.66 to 0.89. The Cronbach's alpha scores for the seven dimensions, intraclass coefficients and effect size estimates were acceptable.

Conclusions: This preliminary evaluation of the IPQ-R HP shows an acceptable to good validity and reliability. Further exploration of the psychometric properties of this questionnaire in a large cohort of healthcare professionals is warranted.

1. Introduction

Illness perceptions are the cognitive beliefs that patients have about their condition. They refer to the cognitive depiction of an illness, reflecting how the illness is 'pictured and stored' in the mind [1,2]. Illness perceptions directly influence the individual's emotional response to the illness and guide coping behavior such as adherence to treatment and health care use in a positive or negative way [1,3,4]. Over time, researchers have used various methodologies to measure the patients' illness perceptions, ranging from questionnaires in early studies to in-depth semi-structured interviews [5]. Unfortunately, these questionnaires were not based on a generally accepted theory nor were they evaluated in different patient groups [6,7].

Currently, the majority of studies focusing on patients' illness perceptions are based upon Leventhal's Self-Regulatory Model. In 1980, Leventhal and colleagues [8] developed this theoretical framework to explain why and how illness representations can differ. They identified different components including the labels, timeline, cause(s), consequences and control. This work resulted in the development of the Illness Perception Questionnaire (IPQ) that assesses these five components of illness representation. A revised version (IPQ-R) was developed in 2002 by Moss-Morris and colleagues [9]. The IPQ-R is widely used in various patient populations such as systemic sclerosis [10] rheumatoid arthritis [11,12], psoriasis [13] and can be modified for use in a particular disease of interest. It has good psychometric properties with a good internal reliability, discrimination and predictive validity and has already been translated in different languages [9].

In daily clinical practice, the patient and the healthcare professional (HP) often have different views on the illness and its impact on a particular patient. Awareness of these divergent illness perceptions is crucial, as they can result in misunderstandings and disrupted communication when unrecognized [14–16]. Previous research evaluating the patient-professional encounter described clear differences between the perceptions of the patients with chronic obstructive pulmonary disease and asthma on the one hand and the physicians and nurse specialists on the other hand concerning timeline, control, consequences and outcomes [17]. Interestingly, a Japanese study [18] demonstrated that a gap between the patient's and the doctor's perceptions was the most significant predictor of doctor-shopping behavior.

Detection of misperceptions is possible by matching a scale that assesses patients' perceptions and perceptions of healthcare professionals (HPs). There is no appropriate and validated instrument available to measure illness perceptions of HPs caring for patients with physical diseases. At this moment questionnaires are available to measure lay perceptions of healthy people [19], illness perceptions of carers of schizophrenia patients [20] and a modified version of the illness perception questionnaire for mental health practitioners[21]. In the latter study the utility of a modified version

of the IPQ was investigated to detect changes in mental health practitioners' illness perceptions about schizophrenia after undertaking psychosocial intervention training. The modified IPQ was completed before and after the training. Afterwards the psychometric properties of the modified IPQ were tested using confirmatory factor analysis showing that a six factor model was most appropriate, but also that there was a poor fit of the items in each factor. This implied that the instrument was not valid and reliable enough to detect changes in illness perceptions.

The purpose of this study was to adapt the IPQ-R to a healthcare professional's version and to perform a preliminary evaluation of its validity and reliability.

2. Methods

The first step in the methodology was an adaptation and rewording of the IPQ-R to a healthcare professionals version. Secondly, face validity and content validity of this adapted version was evaluated in a group of physicians and head nurses. At last, the reliability measurements, i.e. the internal consistency and test-retest reliability, were assessed in a group of nurses.

Adaptation/rewording of the IPQ-R for healthcare professionals

Four authors (SA, PM, JV and RW) discussed and agreed upon the adaptation of the 9-dimension Dutch version of the IPQ-R [22] to a healthcare professionals version (IPQ-R HP). This process comprised several rounds. An item-by-item approach was followed by a dimensional and overall evaluation. The primary goal of the process was to focus on the perception of the HP regarding the disease of a particular patient (dimensions 1, 2, 3, 4 and 6). The secondary focus of the IPQ-R HP was the view of the HP on the perceptions of that particular patient, regarding his/her illness and about its emotional impact on the patient (dimensions 5 and 7). The reformulation of the latter 2 dimensions was done in this way because these dimensions have an emphasis on the emotionality and understanding of the patient and not on the disease of the patient. We did not include the dimensions 'illness identity' (= perceptions of symptoms associated with the illness) and 'causal attributions' in the IPQ-R HP. From a HP's perspective, these 2 dimensions are also part of illness perceptions but more related to medical knowledge or a medical judgment of the illness in comparison with patients' illness perceptions because of their biomedical education. The other 7 dimensions of the original IPQ-R were reworded to a HP's version. Finally, this IPQ-R HP consisted of 7 dimensions: 1) consequences (the HPs' perception of the consequences of the illness for a particular patient); 2) timeline acute/chronic (the HPs' perception about the illness passing quickly or not in a particular patient); 3) personal control (the HPs' perception of the patient's ability to control the illness); 4) treatment control (the HPs' perception about the effectiveness of any treatment or approach to control the illness in a particular patient); 5)

illness coherence (the HPs' perception of the extent to which a particular patient understands their illness); 6) timeline cyclical (the HPs' perception of the cyclical nature of the illness across time); and 7) emotional representations (the HPs' perception of the patients' emotional experience of their illness). In general, besides the rewording and reformulation, the difference between the IPQ-R and IPQ-R HP were the terms 'I' and 'this patient'.

Sampling strategy - Procedure

To measure the face and content validity, a purposive sampling strategy was conducted. We had a list with the names of 20 physicians from 3 general hospitals and 11 head nurses from a large university hospital in Belgium and invited them to participate. Two sampling criteria were used to recruit the healthcare professionals: they had to be specialized in internal medicine and have active patient contact at an outpatient clinic or inpatient service. They were visited in their respective hospitals and introduced in the study (by AVdZ and MVR). Oral and written information about the study was given together with the scoring instructions and the IPQ-R HP. After one week, researchers AVdZ and MVR contacted the physicians and head nurses personally and reminded them to complete the questionnaire if needed.

For the reliability measurement, head nurses of 11 nursing wards from the university hospital were approached and asked if their nurses could participate. At these 11 nursing wards a total of 242 nurses are working. We opted for nurses to score the reliability measurements because for the face and content validity measurements already more physicians than nurses were present. Nurses received first oral information during a team meeting, after which they received written information regarding the study. They were asked to complete the IPQ-R HP on the basis of 4 patient vignettes. After an interval of 4 weeks, they were asked to complete the IPQ-R HP on the basis of 1 patient vignette that was included in the first round. The reason for reducing the number of vignettes from 4 to 1 was the indication of survey fatigue among respondents which could have an impact on the response rate. These patient vignettes were developed by SA and RW and comprised information regarding 2 patients with systemic lupus erythematosus and 2 patients with systemic sclerosis based on real patients seen in the clinic. The information in the vignettes pertained to the patients' clinical condition (i.e. a description of antibody profile, characteristics and complication of the disease), the medical treatment and eventual psychosocial complications and coping styles having a possible or probable impact on daily life.

Validity

Face Validity

Face validity is the extent to which a test is representative for covering the concept it purports to measure at first sight [23]. The IPQ-R HP was accompanied by four questions about each dimension and also a general question at the end. These four questions were: “Are these questions a correct representation of the dimension?”; “Are the questions clear?”, “Are there questions lacking?” and “Are there redundant questions?” At last, there was an open question for further remarks. The reason why we asked, if the items per dimension are representative for a particular dimension, is because the concept of illness perceptions consists of several dimensions. So, we used the theory behind the concept of illness perceptions [8] as a backbone to rely on. In this phase, the emphasis was on the representativeness of the items covering the concept on first sight and not on removing or adding new items because of their content [24].

Content validity

The IPQ-R HP was also tested for content validity. Content validity is the extent to which a measure represents all facets of a given construct [25]. In other words, the items on the test represent the entire range of possible items the test should cover [26,27] .

Physicians and head nurses, this was the same group as for the appraisal of the face validity, were instructed to rate the 38 items of the IPQ-R HP on a 4-point Likert scale as: “1 = not relevant”, “2= somewhat relevant”, “3= quite relevant”, “4= highly relevant”. An appropriate sample size for calculating content validity ranges between 5 and 10 [26].

Based on these data, the item Content Validity Index (I-CVI) was calculated. The I-CVI is the proportion of items that received a rating of 3 or 4 by the experts. For the total instrument and each scale, a scale content validity index (S-CVI) was calculated. This is the average of all the I-CVI’s of the individual items ($S\text{-}CVI_{ave}$). An I-CVI of 0.78 and an $S\text{-}CVI_{ave}$ of 0.90 is considered to be excellent [27] .

To counter the limitations of the CVI, each I-CVI was adjusted for chance agreement by calculating the modified kappa statistic (k*) [28]. To compute the modified kappa, the probability of chance agreement was computed first: $P_c = [N!/A!\,(N-A)!] \times 0.5^N$ where N is the number of experts and A is the number agreeing on good relevance (rating 3 and 4). Next, the k* was calculated with the formula $k^* = [I\text{-}CVI - P_c]/[1 - P_c]$ [26]. According to the standards of Fleiss (2003) [29] and Cicchetti and Sparrow (1981) [30] the value of each k* was evaluated as poor (k <0.40), fair (k of 0.40 to 0.59), good (k of 0.60 to 0.74), or excellent (k of > 0.74).

Reliability

To measure the internal consistency, Cronbach's alpha values per vignette were calculated and also a total Cronbach's alpha value was computed. Sample size estimations to accurately determine the internal consistency showed that a minimum of 17 subjects was necessary.

To evaluate the test-retest reliability, nurses were asked at time point 1 (T1) to complete the 4 vignettes and at time point 2 (T2) they were asked to complete vignette number 4 because this vignette had the best alpha values and the content was a good mix of psychosocial and clinical information. Intraclass correlation (ICC) was computed to describe how strongly illness perception dimensions in the same group resemble each other. It is a measure of the reliability of measurements. ICC can be interpreted as follows: 0-0.2= poor agreement; 0.3-0.4= fair agreement; 0.5-0.6= moderate agreement; 0.7-0.8= strong agreement; and >0.8= almost perfect agreement. These values are arbitrary cutoffs, but similar to those used by Landis and Koch [31] for agreement of categorical items.

We also looked for differences in illness perception scores between T1 and T2 which are expressed in effect sizes. For the continuous variables, an effect size for the Wilcoxon signed rank test was calculated by $r = \frac{Z}{\sqrt{N}}$ where Z is the normal approximation of the Wilcoxon test statistic and N is the total number of participants on which Z is based. To appraise the magnitude of the effect sizes we used the cutoff values for Cohen's r: small effect size= between 0.10 and 0.30; medium effect size= between 0.30 and 0.50 and large effect size= 0.50 or higher [32].

Statistical analysis

Descriptive statistics were used to calculate the I-CVI values, the $S\text{-}CVI_{ave}$ values, the P_c and k*. These data were analysed using Microsoft Excel (version 2011). The calculation of the Cronbach's alpha, ICC and Wilcoxon signed rank test was carried out with SPSS version 22.0.

Ethics approval and consent to participate

By answering the questions and completing the questionnaire concerning face and content validity potential respondents (physicians and head nurses) gave their consent to participate in the study. Participation was voluntary and questionnaires were treated anonymously.

For the construction of the patient vignettes and completion of the IPQ-R HP on the basis of the vignettes, ethical approval by the Institutional Review Board of the University Hospitals Leuven was received (study number S57719). Only the patient vignettes of the patients who gave permission to put their medical and psychosocial data in the format of a vignette, were included. Nurses, who had to complete the IPQ-R HP on the basis of the patient vignettes, were included after they gave written informed consent.

3. Results

Experts scoring validity

Seventeen doctors and 9 head nurses participated (response rate = 84%). The sample of the physicians consisted of 9 men and 8 women and was composed of 4 gastroenterologists; 3 endocrinologists; 2 rheumatologists; 2 cardiologists; 2 pulmonologists; 1 nephrologist; 1 neurologist; 1 dermatologist and 1 oncologist. No further demographic data were available. The 3 doctors who did not participate were all men and gave a lack of time as a reason for not participating. The 9 head nurses, 6 women and 3 men, worked at following disciplines: cardiology; gastroenterology; rheumatology; nephrology; gynecology; ophthalmology; otorhinolaryngology and 2 pulmonology wards. One head nurse declined participation because of insecurity concerning scoring the questions correctly. The other gave no reason.

Face validity

In Table 1, the face validity scores of the IPQ-R HP are tabulated. One of the 9 head nurses did not score the face validity questions. For almost all healthcare professionals, the questions were a correct representation of the dimensions and the questions were clear. Physicians wanted to add more questions such as items concerning self-appearance, autonomy and quality of life in the 'Consequences' dimension. In the 'Timeline' dimension they wanted to add questions regarding curability of the patient and worsening of the disease. The most redundant or overlapping questions for the physicians and head nurses were found in the dimensions Timeline acute/chronic; Personal control; Illness coherence and Emotional representations. More specifically, for the dimension 'timeline acute/chronic', the experts found an overlap in items 7, 8, 10 & 11. For the dimension 'personal control', following items were comparable for the experts: 14, 15, 17, 18. For the dimension 'illness coherence', the experts scored items 25, 26, 27 as comparable items. For the dimension 'emotional representations', items 37 & 38 were comparable.

Table 1: Overview appraisal face-validity of the IPQ-R HP

Dimensions IPQ-R HP	Are the questions a correct representation of the dimension?		Are the questions clear?		Are there questions lacking?		Are there questions redundant?	
	Doctors n=17	Nurses n= 8	Doctors n= 17	Nurses n=8	Doctors n= 17	Nurses n=8	Doctors n= 17	Nurses n=8
Consequences	17/17	8/8	17/17	6/8	4/17	1/8	6/17	1/8
Timeline acute/chronic	16/17	8/8	15/17	8/8	4/17	0/8	7/17	4/8
Personal Control	14/17	8/8	14/17	8/8	2/17	0/8	8/17	3/8
Treatment Control	15/17	8/8	14/17	7/8	1/17	0/8	2/17	2/8
Illness Coherence	15/17	8/8	12/17	7/8	1/17	0/8	7/17	4/8
Timeline Cyclical	15/17	8/8	15/17	8/8	3/17	0/8	4/17	2/8
Emotional Representations	15/17	8/8	15/17	8/8	2/17	0/8	12/17	4/8

Content validity

A total of 16 physicians and 9 head nurses completed the 4-point Likert scale (see Table 2). Three doctors did not complete one question or one dimension. They gave no reason why they left these items blank. This means that 12 items were rated by 15 doctors and 26 items were assessed by 16 doctors.

Twenty-eight of the 38 items had an excellent content validity (I-CVI ≥ 0.78 and k* > 0.74), 4 of the 38 items had a good content validity (I-CVI <0.78 and 0.60 ≤ k* ≤ 0.74) and 4 of the 38 items had a fair content validity (I-CVI <0.78 and 0.40 ≤ k* ≤ 0.59) (see Table 1). Two items (item 4 and item 10) had a very low modified kappa (k*< 0.40) and were considered content invalid.

The average scale content validity ($S\text{-}CVI_{Ave}$) for each of the 7 dimensions was as follows: Consequences was 0.75; Timeline acute/chronic was 0.75; Personal control was 0.81; Treatment control was 0.89; Illness coherence was 0.74; Timeline cyclical was 0.66; and Emotional representations was 0.77. The $S\text{-}CVI_{Ave}$ for the entire questionnaire was 0.79.

After omitting items with a fair and very low k* value the $S\text{-}CVI_{Ave}$ for Consequences was 0.88 (without item 3 and 4), for Timeline acute/chronic was 0.83 (without item 10), Illness coherence was 0.88 (without item 25 and 27) and Emotional representations was 0.80 (without item 34). The $S\text{-}CVI_{Ave}$ for the entire questionnaire after removing items 3, 4, 10, 25, 27 and 34 was 0.82.

For the group of the nurses, very low modified kappa values (k*< 0.40) were present for item 3 (k*= 0.26), 10 (k*=0.09) and 27 (k*=0.26). Item 4 had a k* value of 0.42.

Table 2: Evaluation of the content validity of the IPQ-R HP

	Item (dimension)	Experts (n)	Experts with rating 3 or 4	I-CVI[a]	P_c[b]	k*[c]	Evaluation[d]
1	The illness of my patient is serious (1)	25	23	0.92	0.0000	0.92	Excellent
2	The illness of my patient has major consequences on his/her life (1)	25	25	1.00	0.0000	1.00	Excellent
3	The illness of my patient does not have much effect on his/her life (1)	25	13	0.52	0.1550	0.43	Fair
4	The illness of my patient strongly affects the way others see him/her (1)	25	12	0.48	0.1550	0.38	Poor
5	The illness of my patient has serious financial consequences for him/her (1)	25	18	0.72	0.0143	0.72	Good
6	The illness of my patient causes difficulties for those who are close to him/her (1)	24	21	0.88	0.0001	0.87	Excellent
7	The illness of my patient will last a short time (2)	25	17	0.68	0.0322	0.67	Good
8	The illness of my patient is likely to be permanent rather than temporary (2)	25	24	0.96	0.0000	0.96	Excellent
9	The illness of my patient will last for a long time (2)	25	21	0.84	0.0004	0.84	Excellent
10	The illness of my patient will pass quickly (2)	25	8	0.32	0.0322	0.30	Poor
11	I expect that my patient will have his/her illness for the rest of his/her life (2)	24	19	0.79	0.0025	0.79	Excellent
12	The illness of my patient will improve in time (2)	25	22	0.88	0.0001	0.88	Excellent
13	My patient can do a lot to control his/her symptoms (3)	25	22	0.88	0.0001	0.88	Excellent
14	What my patient does can determine whether his/her illness gets better or worse (3)	25	21	0.84	0.0004	0.84	Excellent
15	The course of my patients' illness depends on him/her (3)	25	21	0.84	0.0004	0.84	Excellent
16	Nothing my patient does will affect his/her illness (3)	25	19	0.76	0.0053	0.76	Excellent
17	My patient has the power to influence his/her illness (3)	25	18	0.72	0.0143	0.72	Good
18	The actions of my patient will have no effect on the outcome of his/her illness (3)	25	21	0.84	0.0004	0.84	Excellent
19	There is very little that can be done to improve the illness of my patient (4)	25	22	0.88	0.0001	0.88	Excellent
20	The treatment of my patient, will be effective in curing his/her illness (4)	25	23	0.92	0.0000	0.92	Excellent
21	The negative effects of the illness of my patient can be prevented (avoided) by his/her treatment (4)	25	23	0.92	0.0000	0.92	Excellent
22	The treatment of my patient can control his/her illness (4)	25	25	1.00	0.0000	1.00	Excellent
23	There is nothing which can help the condition of my patient (4)	25	18	0.72	0.0143	0.72	Good
24	The illness of my patient is a mystery to him/her (5)	25	23	0.92	0.0000	0.92	Excellent
25	The symptoms of the condition of my patient are puzzling to him/her (5)	25	14	0.56	0.1328	0.49	Fair
26	My patient does not understand his/her illness (5)	25	20	0.80	0.0016	0.80	Excellent
27	The illness of my patient doesn't make any sense to him/her (5)	25	13	0.52	0.1550	0.43	Fair
28	My patient has a clear picture or understanding of his/her condition (5)	25	23	0.92	0.0000	0.92	Excellent

29	The symptoms of the illness of my patient change a great deal from day to day (6)	24	19	0.79	0.0025	0.79	Excellent
30	The symptoms of my patient come and go in cycles (6)	24	18	0.75	0.0080	0.75	Excellent
31	The illness of my patient is very unpredictable (6)	24	22	0.92	0.0000	0.92	Excellent
32	My patient goes through cycles in which his/her illness gets better and worse (6)	24	20	0.83	0.0006	0.83	Excellent
33	My patient gets depressed when he/she thinks about his/her illness (7)	24	20	0.83	0.0006	0.83	Excellent
34	When my patient thinks about his/her illness he/she gets upset (7)	24	15	0.63	0.0779	0.59	Fair
35	The illness of my patient makes him angry (7)	24	19	0.79	0.0025	0.79	Excellent
36	The illness of my patient does worry him/her (7)	24	19	0.79	0.0025	0.79	Excellent
37	Having this illness makes my patient feel anxious (7)	24	19	0.79	0.0025	0.79	Excellent
38	The illness of my patient makes him/her afraid (7)	24	19	0.79	0.0025	0.79	Excellent

[a] I-CVI (item content validity index) = number giving a rating of 3 or 4 / number experts

[b] p_c (probability of chance occurrence) = $[N!/ A!(N-A)!] \times 0,5^N$ where N = number of experts and A = number agreeing on good relevance

[c] k^* = kappa designating agreement on relevance: $k^* = (I\text{-}CVI - p_c) / (1 - p_c)$

[d] Evaluation criteria for k^*: poor = $k^* < 0.40$; fair = k^* of 0.40-0.59; good = k^* of 0.60-0.74; excellent = $k^* > 0.74$

Experts scoring reliability

Four head nurses of following wards agreed with the participation of their nurses: pulmonology, rheumatology, nephrology and internal medicine. A total of 20 nurses gave consent for participation, comprising 15 women and 5 men, with a mean age of 39 years (SD= 12) and mean years of working experience of 17 years (SD= 12). Information about the non-responders is not available.

Internal consistency

Depending on the sample size estimations, a sample of 20 nurses was large enough to adequately compute the Cronbach's alpha. Total Cronbach's alpha values on all vignettes rated by 20 nurses are: Consequences= 0.78; Timeline acute-chronic= 0.77; Personal control = 0.80; Treatment control = 0.50; Illness coherence = 0.75; Timeline cyclical = 0.80; Emotional representations= 0.86 (see Table 3).

Table 3: Cronbach's alpha values for the four patient vignettes

Dimensions **IPQ-R HP**	**Alpha value** Vignette 1	**Alpha value** Vignette 2	**Alpha value** Vignette 3	**Alpha value** Vignette 4	**Alpha value** All vignettes
Consequences	0.77	0.81	0.73	0.83	0.78
Timeline acute-chronic	0.89	0.67	0.78	0.75	0.77
Personal control	0.80	0.88	0.77	0.76	0.80
Treatment control	0.48	0.66	0.30	0.54	0.50
Illness coherence	0.77	0.63	0.75	0.86	0.75
Timeline cyclical	0.80	0.72	0.84	0.84	0.80
Emotional representations	0.86	0.85	0.93	0.81	0.86

Test-retest reliability

Thirteen nurses completed the IPQ-R HP on the basis of vignette 4 at T2. Table 4 displays the ICC values and Wilcoxon z-score with effect size calculation for all dimensions. The ICC values were strong for all dimensions, except for Personal control (ICC= 0.444) and Timeline cyclical (ICC= 0.417). For the latter two dimensions the ICC values can be considered as fair which means that there is a fair agreement of Personal control and Timeline cyclical at the two time points. The effect size estimates between the 2 moments was small, which means that differences in the scores of the nurses between the two time points were small.

Table 4: Test-retest intraclass correlation and Wilcoxon signed rank test

IPQ-R HP subscales	4 weeks test-retest intraclass correlation (95% CI)	4 weeks test-retest Wilcoxon z score (effect size*)
Consequences	0.866 (0.618-0.957)	-1.221 (-0.24))
Timeline acute/chronic	0.751 (0.364-0.917)	-0.938 (-0.18)
Personal control	0.444 (-0.116-0.790)	-0.237 (-0.05)
Treatment control	0.839 (0.553-0.948)	-0.289 (-0.06)
Illness coherence	0.713 (0.264-0.908)	-1.086 (-0.21)
Timeline cyclical	0.417 (-0.148-0.777)	-0.843 (-0.17)
Emotional representations	0.844 (0.567-0.950)	-1.370 (-0.27)

* Effect size: small = between 0.10 and 0.30; medium = between 0.30 and 0.50; large = 0.50 or higher

4. Discussion

The purpose of this study was to adapt and to perform a preliminary evaluation of the validity and reliability of the IPQ-R HP. At first sight, this IPQ-R HP has a good and acceptable face and content validity, and reliability. Experts judged the majority of the items as relevant. Item 4 and 10 were the only items with a poor or very low kappa value, indicating that these items are not valid to measure the construct, i.e. illness perceptions. Nonetheless, we decided to keep all items in the IPQ-R HP and did not omit item 4 and 10. The reason why we did not delete item 4 and 10 was that these low scores are probably due to the fact that a mix of professions, nurses and physicians, scored these items which means that they maybe gave a different meaning or interpretation to this. Only a confirmatory factor analysis can give information about items that certainly should be omitted.

The internal consistency of the 7 dimensions was acceptable and the instrument had overall good scores for the reliability measurements except for the treatment control dimension. The treatment control dimension with an alpha value of 0.50 (calculated for all vignettes) was the lowest in comparison with the other dimensions. Literature [33] states that possible reasons for a low value of alpha could be a low number of questions, poor interrelatedness between items or heterogeneous constructs. Therefore, we think that in our study a combination of a low number of items -namely, 5 items- and a low interrelatedness of these items are possible reasons why the treatment control dimension has the lowest alpha value in comparison with the other dimensions. On the other hand, experts in our study had the opinion that the items of the treatment control dimension were representative for this dimension at first sight and they also scored the content validity of this dimension as excellent. Probably a confirmatory factor analysis in a large sample of HPs can give us

more insight. By comparison of the alpha values of the study of Fleming et al. (2009) [21] with the total alpha values of our study, we found that our alpha scores were higher. A possible reason is that in our study at least 4 items per dimension are present. The study of Fleming et al. (2009) [21]had 2 items per dimension. Fleming et al. (2009) [21] not only calculated Cronbach's alpha values, but went a step further and used factor analysis to determine the underlying structure of the IPQ that they modified. The authors stated that a six factor model was the most appropriate model in comparison with a five factor model or one-dimensional model. However, no extra information was given about the p-values or correlations between the items and construct (factor) in the model. Their shortcoming was also the limited number of items (2) per factor leading to a non-representative result. A strength was the sample size of 245 mental health practitioners which was sufficient for this kind of analysis.

The strengths of our study are that we conducted this research in a group of physicians, head nurses and nurses employed in different medical disciplines and four hospitals. The sample size to measure the content validity was also much larger than previously used in similar studies [26]. The high response rate was probably due to the personal contact that we had before conducting the study. Another strength is that almost all physicians and head nurses considered the questions to be clear and providing a correct representation of the dimension at first sight. We were able to keep the original construction of the questions, which allows for matching with patients' questionnaires at individual item level. The method of Lynn [26] is considered as an extensive method to evaluate the content validity and has shown valuable results. The results of both measurements, I-CVI and k* were in line with each other, with items not meeting the I-CVI criterion of 0.78 not showing excellent k* values and vice versa, indicating that both methods resulted in the same conclusion and were strengthening current evidence. For the reliability analyses we calculated the alpha value for each vignette separately. This gave us an idea about the amount of influence of the quality of the vignette on the reliability estimate and revealed that vignette 4 had the best alpha values.

Shortcomings of this study concerning the validity measurements, is that the use of cognitive interviewing techniques asking physicians and head nurses about their reflections concerning the individual items would, have given more background information about questions that were not clear or were skipped. Another shortcoming was that the sample size was not large enough to compute a confirmatory factor analysis because we needed then a sample between 380-570 healthcare professionals (i.e. 10-15 respondents per item) [34]. A confirmatory factor analysis would give information regarding the unidimensionality of the subscales and also provides information about the relationship between each item and the subscale. For the reliability measurements, it was difficult to motivate the nurses to complete the 4 vignettes and it was even much more difficult to motivate them to complete one vignette for a second time. Therefore, we used vignette 4 for the retest, which was a good mix of clinical and psychosocial information and had also the highest alpha value. This could have

led to an overestimation of the intraclass coefficient because we used the vignette with the highest interrelatedness of items. Reasons for the low response rate were no time, too many questionnaires and too difficult vignettes. For planning further research, the number of the vignettes have to be taken into consideration.

Another limitation was that the reliability estimates are based just on a sample of nurses. It is unclear whether these results are generalizable to physicians because nurses and physicians differ in a variety of aspects like education, patient contact, responsibility for diagnostics and treatment. As a last point to consider we want to mention that in the adaptation process of the IPQ-R to the IPQ-R HP we omitted the identity and causality dimensions of illness representations. Our reasoning was that these 2 dimensions are –in comparison with patients' perceptions- more related with biomedical knowledge. A remark on this is on the one hand, that treatment decisions are often based on physicians' representations of the identity and causal attributions dimensions and on the other hand it is possible that conflicts between patients and physicians arise when they differ in their opinions about which symptoms relate to a specific illness or which factors caused a particular disease.

After a comprehensive validation process, we can explore the potential applications of this questionnaire in patient care. This tool is useful for investigating the causes of misunderstandings and conflicts that have arisen between medical staff and patients. When differences in perceptions between patients and HPs are detected than these differences can be discussed using this tool by comparing the patient's and HP's version with each other. In this way, HPs can reflect upon their own beliefs and how much it differs from patients' beliefs. When HPs are aware of these differences they can work in a patient-centered manner during patient education sessions which means that some items or some dimensions can be a stepping stone to tailor information for a particular patient. With the IPQ-R HP areas of disagreement between patients' and HPs' perceptions can be pinpointed in a more detailed way which is an advantage because in this way the communication and shared understanding between HPs and patients can be enhanced. This is important because doctor–patient communication is a powerful indicator to achieve quality in care determining patients' self-management behavior and ultimately health outcomes [35,36].

Practically, the patient can complete their version in the waiting room- this means before the doctor has seen him/her- and the doctor or other HP can complete the IPQ-R HP after the patient's visit. These questionnaires can be completed in every setting, i.e. an inpatient or outpatient setting. It is important that this happens when the HP has formed an idea about the patient's physical and mental condition. The next stage is the comparison of these 2 instruments which can be done easily by the HP. We do not think it is useful to complete this questionnaire each moment the patient encounters an HP. The completion of these instruments can have an added value especially at diagnosis and when

there is a flare or acute exacerbation of the patient's condition because illness perceptions are relatively stable but may show some fluctuations at those time points [37].

In **conclusion,** the IPQ-R HP appears adequate and useful to assess the perception of healthcare professionals concerning the illness of an individual patient and produces -in this preliminary phase- reliable and valid output. A more extensive validation process is needed in a large cohort of healthcare professionals to explore the psychometric properties of this questionnaire prior to a widespread use in clinical practice. Moreover, a large cohort of healthcare professionals is needed to investigate the factor structure of the IPQ-R HP with the aim to determine which of the items best represent each of the illness perception dimensions. In this way it is possible to have more insight in the construct validity of the IPQ-R HP.

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4

DIVERGING ILLNESS PERCEPTIONS BETWEEN PHYSICIANS ABOUT PATIENTS WITH SYSTEMIC LUPUS ERYTHEMATOSUS AND SYSTEMIC SCLEROSIS: A VIGNETTE-BASED STUDY

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Abstract

Objective: Systemic Lupus Erythematosus (SLE) and Systemic Sclerosis (SSc) are complex chronic auto-immune diseases characterized by multiple organ involvement, comorbidities and complications. This complexity results in a need for a multidisciplinary management and treatment of SLE and SSc by physicians from a number of medical disciplines, all of who may have different perceptions concerning the condition of a particular patient. The aim of this study was to explore differences in physicians' perceptions on the illness of SLE and SSc patients.

Methods: Physicians from nine disciplines working at three hospitals in Belgium completed illness perception questionnaires for healthcare professionals based on 4 patient vignettes, i.e. 2 vignettes per disease (SLE-SSc). Statistical analysis was carried out by a k-means clustering technique for clustering physicians according to their illness perceptions.

Results: Fifty physicians, 62% men with a mean age of 42.8 years (SD =11.3) and mean working experience of 12.7 years (SD=11.6), participated. For each disease, three clusters of physicians with different scores in illness perceptions were identified. For SLE, these clusters were specified as the 'optimistic' group, the 'realistic' group and the 'overwhelming impact by disease' group. For SSc, the clusters were characterized as the 'optimistic' group, the 'realistic' group and the 'skeptical' group.

Conclusions: We found divergent illness perceptions across physicians of the same and other disciplines. Our study yielded three clusters of physicians per disease with a large variability in illness perceptions. Further studies should focus on the factors that determine these differences and their consequences for patient care.

1. Introduction

Systemic lupus erythematosus (SLE) and systemic sclerosis (SSc) are chronic systemic auto-immune diseases characterized by multiple organ involvement and many related morbidities. For example, SSc patients experience a high disease burden with potential digital ulcers, dyspnea, fecal incontinence, and erectile dysfunction [1,2]. While, in SLE, Raynaud's phenomenon, skin, kidney, and cerebral involvement might occur and SLE patients are at risk of cardiovascular complications and avascular necrosis. An eventual pregnancy poses specific challenges [3].

In practice, patients are treated by often prolonged and complex immunosuppressive strategies with regular monitoring. Yet, these diseases are driven by often insufficiently controlled inflammation and it is clear that the complexity of multiple organ involvement, comorbidities and complications implies that the treatment approach needs to be multidimensional by relieving symptoms, reducing and preventing organ dysfunction and slow down disease progression [4]. Moreover, besides the specific organ impact, literature shows that these diseases also have a considerable impact on the health-related quality of life of patients and their ability to carry out activities at home and at work due to for instance fatigue [5], pain (from digital ulcers, musculoskeletal pain, etc.) [6], decreased physical functioning in upper and lower extremities (caused by contractures, joint pain, skin tightness, Raynaud's phenomenon, etc.) [7–10].

Thus, not only the drug treatment but also the global management of these two potentially devastating chronic diseases needs monitoring by a multidisciplinary team [11]. A multidisciplinary care team for SLE and SSc patients should involve healthcare professionals from different professions and different specialties, including physicians, specialist nurses, physical therapists, occupational therapists, psychologists, and social workers. The coordination of the care team may fall to a dedicated specialist, often a rheumatologist, who will act as the clinical architect, coordinating the care and referring the patient to and working together with multiple specialists of other disciplines who deal with organ-specific manifestations, comorbidities or complications [12].

However, physicians may hold certain views or perceptions about the condition of their patients, the disease characteristics, the treatment, and the prognosis, which, amongst other things, may be based on eminence or personal experience in daily clinical practice, on the frequency of patient contact, or on a broader focus of the physician than on only disease-specific characteristics such as an additional focus on quality of life and societal participation. Hence, a rheumatologist, who coordinates the care for systemic diseases, might perceive the illness of a patient with SLE and SSc in a different way compared to colleagues from other medical disciplines or colleagues of the same discipline involved in a multidisciplinary approach to care.

Such illness perceptions of physicians which are relatively enduring cognitive schemes that stimulate and guide action are important because on the one hand doctors' explanations will affect patients' thinking about their condition [13], and on the other hand differences in perceptions regarding the illness of a patient could adversely affect patient care or cause inconsistent advice, confusions, and misconceptions amongst patients and physicians.

Despite the likely relevance of doctors' illness perceptions for the management of patients with rheumatic conditions, relatively few studies have addressed this topic. One UK study [14] in rheumatoid hand surgery, detected differences in perceptions, related to the main indications and expected results for surgery between rheumatologists, surgeons, and therapists in every aspect of the procedure of management of the hand. Evidence from other disease specialties such as epilepsy and nonepileptic seizures describes [15] similar discrepancies between specialists from different disciplines. Neurologists perceived epilepsy as a more chronic condition and having a less emotional impact for the patient in comparison with psychiatrists. In the case of nonepileptic seizures, psychiatrists perceived less negative consequences of this condition for the patient than the neurologists do. These studies demonstrate the potential for problems with communication, treatment, and outcome, which might arise as patients are referred from one specialty to the other.

Currently, no evidence on differences in perceptions of systemic diseases between physicians from several disciplines is available. For this purpose, we sought to gather more insight in the perceptions of different medical specialists concerning the illness of a patient with SLE and SSc. The aim of this study was to explore differences in physicians' perceptions on the illness of patients with SLE and SSc in a current practice setting in Belgium.

2. Methods

Procedure

Until now, patient vignettes were mostly used in the literature for measuring clinical decision making and the efficiency of clinical care [16,17]. In this study, we used patient vignettes, as a novel technique to get more insight in the cognitive and emotional representations of physicians about a particular patient with SLE or SSc.

Four patients—two patients with SLE and two patients with SSc—who are currently followed in our outpatient clinic, were selected by two researchers (SA and RW) based on their multiple organ involvement and comorbidities. Vignettes, which focused on the whole person with a disease, were constructed based on these patients' clinical condition, complications, antibody profile, the current and previous medical treatment, and the psychosocial situation. The accuracy and correctness of each

vignette was separately reviewed by the respective patients whose names were changed for anonymity reasons. A description of the vignettes is available as an additional file (see Appendix 1).

Between October 24, 2014 and January 31, 2015, physicians employed in the largest university hospital and two of the largest general hospitals in Belgium were approached for participation. The physicians approached were from nine medical disciplines: rheumatology, cardiology, pulmonology, gynecology, internal medicine, nephrology, ophthalmology, dermatology, and gastroenterology. Postal questionnaires and 4 patient vignettes together with a pre-stamped return envelope were sent to physicians. Electronic reminders were sent 14 and 28 days after sending the questionnaires.

The inclusion criteria were that physicians had to be certified in their specialty or in the last 2 years of their specialist training, and had a former experience with SLE or SSc patients. We choose a convenience sampling technique because many different physicians may take an interest in the care for patients with systemic diseases.

Measures

Demographic questionnaire

Age, gender, frequency of contact with SLE or SSc patients, years of working experience and hospital of employment were recorded.

Revised Illness Perception Questionnaire for Healthcare Professionals (IPQ-R HP)

Each physician was asked to read each of the four vignettes and to answer the IPQ-R HP that accompanied each vignette. The IPQ-R HP [18]is an adapted version of the IPQ-R [19] for healthcare professionals (HP). The IPQ-R HP is a 38-item questionnaire rated on a 5-point Likert scale (1 = “strongly disagree”—5 = “strongly agree”) and captures seven dimensions: (1) consequences of the illness for a particular patient as assessed by the HP; (2) timeline acute/chronic: the HP’s perception about the illness passing quickly or not in a particular patient; (3) personal control: the HP’s perception of the patient’s ability to control the illness; (4) treatment control: the HP’s perceptions about the effectiveness of any treatment or approach to control the illness in a particular patient; (5) illness coherence: the HP’s perception of the extent to which a particular patient understand his/her illness; (6) timeline cyclical: the HP’s perception of the cyclical nature of the illness across time; and (7) Emotional representations: the HP’s perception of the patient’s emotional experience of their illness.

The IPQ-R HP has an acceptable to good face and content validity, internal consistency, and test–retest reliability [18].

Statistical analysis

Descriptive analyses were carried out with Excel version 2010. A k-means clustering technique was performed to identify groups of physicians according to their illness perceptions [20,21]. Prior to cluster analysis, the mean z-scores of the seven illness perception dimensions of the IPQ-R HP were calculated.

The process begins by choosing k observations to serve as the center of each proposed cluster. Then, the distance from each of the other observations is calculated for each of the k clusters, and observations are put in the cluster to which they are the closest. After each observation has been assigned to a cluster, the center of the clusters is recalculated, and every observation is checked to see if it might be closer to a different cluster. This is an iterative process until convergence. The cluster analysis was conducted separately for both diseases and was carried out with SAS version 9.4. The further labeling of the clusters was based on thematic analysis. This means that patterns across data were identified, analyzed, and reported by two authors (SA and RW).

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Ethical approval was obtained from the Institutional Review Board of the University Hospital and the local ethics committee of the participating centers (IDnr. B322201421473).

3. Results

Descriptive analysis

Questionnaires were sent to 284 physicians of whom 81 responded. Compared to non-responders, a greater proportion of those who responded were male (63.3 vs 50.3%). No other data are available. Of these 81 physicians, 50 were eligible for participation in the study (see Fig.1).

Flow diagram of inclusion procedure

The demographic data of the 50 participating physicians and their distribution per discipline are presented in Table1 Ten physicians were in the last 2 years of their specialist training. The sample consisted of 31 men and 19 women with a mean age of 42.8 years (SD 11.3) and a mean working experience of 12.7 years (SD 11.6). Nine physicians (19.2%) had contact with SLE or SSc patients several times a week and 16 patients (34%) several times a year. A total of 13 rheumatologists were employed at the three hospitals, of those, 5 (38%) took part in the study.

Figure 1: flow diagram of inclusion procedure

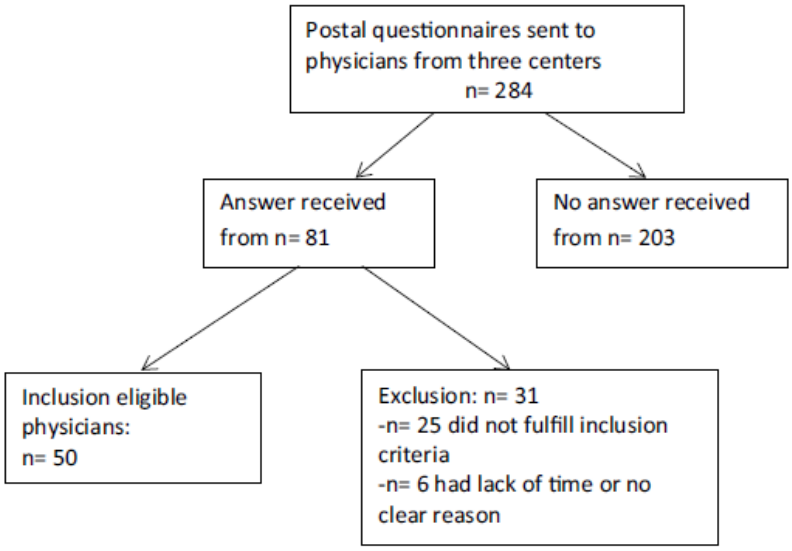


Table 1: Demographics and distribution of all participating physicians

All medical specialists	**N= 50**
Gender (n)	
Men	31 (62.0%)
Women	19 (38.0%)
Age (in years), mean +/- SD	42.8 +/-11.3
Work experience (in years)*, mean +/- SD	12.7+/-11.6
Frequency of contact with SLE or SSc patients ° (%)	
Several times a week	9 (19.2%)
Once a week	6 (12.8%)
Several times a month	5 (10.6%)
Once a month	8 (17.0%)
Several times a year	16 (34.0%)
Once a year	3 (6.4%)
Hospital (n)	
University Hospital	26 (52.0%)
General hospital 1	10 (20.0%)
General hospital 2	14 (28.0%)
Distribution per discipline (n)	
Rheumatology	5 (10.0%)
Cardiology	4 (8.0%)
Pulmonology	2 (4.0%)
Nephrology	11(22.0%)
Gastroenterology	5 (10.0%)
Internal medicine	6 (12.0%)
Dermatology	5 (10.0%)
Ophthalmology	7 (14.0%)
Gynaecology	5 (10.0%)

*calculated on n= 40; °calculated on n= 47

Main analysis

Description of the clusters

Cluster analysis revealed three clusters of physicians' perceptions per disease (SLE-SSc). The mean z-scores of the illness perception dimensions of the three obtained clusters per disease are shown in Figs. 2 and 3, respectively.

Mean z-scores of the illness perception dimensions per cluster for SSc

For SLE, the physicians in cluster 1 (42%) reported less severe consequences of the disease for the patient, less emotional impact and believed that the disease was clearly understood by the patient. They also perceived the time course of SLE as more acute and less recurrent and had the opinion that the patient herself and the treatment can control the illness. Cluster 1 was called the 'optimistic' group. Cluster 2 (36%), reported more severe consequences of the disease for the patient and a more chronic, and cyclical disease course. These physicians perceived SLE as controllable by the patient but less so by treatment. They also reported an emotional impact of SLE on the patient and less understanding of SLE by the patient. This cluster was specified as the 'realistic' group. Cluster 3 (22%) reported less severe consequences of SLE for the patient and a more acute and less recurrent disease course. These physicians perceived SLE as less controllable by the patient and by the treatment and had the opinion that the patient does not fully understand her illness. Furthermore, they perceived a high emotional impact of SLE for the patient, which was the reason why we identified this cluster as the 'overwhelming impact of disease' group.

For SSc, cluster 1 (22%) comprised the physicians who reported less severe consequences due to SSc for the patient; a time course that was perceived as acute but recurrent; less emotional impact of the illness. They also felt that the condition could be controlled by the patient and by treatment and held the opinion that the patient has a clear understanding of SSc. To this end, we specified this cluster as the 'optimistic' group. Cluster 2 (42%), reported more severe consequences for the patient due to SSc. Physicians believed that SSc had a chronic time course with a cyclical nature and somewhat believed that it could be controlled by the patient and by the treatment. They also believed that patients understood their SSc. Cluster 2 was called the 'realistic' cluster. Cluster 3 (36%) describes the perception of some consequences of SSc for the patient, the chronicity and noncyclical time course; the perception that the patient has less personal control and less control by treatment; a less emotional patient with some understanding of SSc. Cluster 3 was labeled as the 'skeptical group'.

Cross-tabulation of cluster membership between the diseases showed that 15 physicians who were assigned to the SLE realistic group were also in the SSc realistic, suggesting good correspondence between the physicians in both disease groups. Just eight physicians of those in the SLE optimistic

group were also present in the SSc optimistic group, suggesting little concordance between these groups.

Figure 2: Mean z-scores of the illness perception dimensions per cluster for SLE

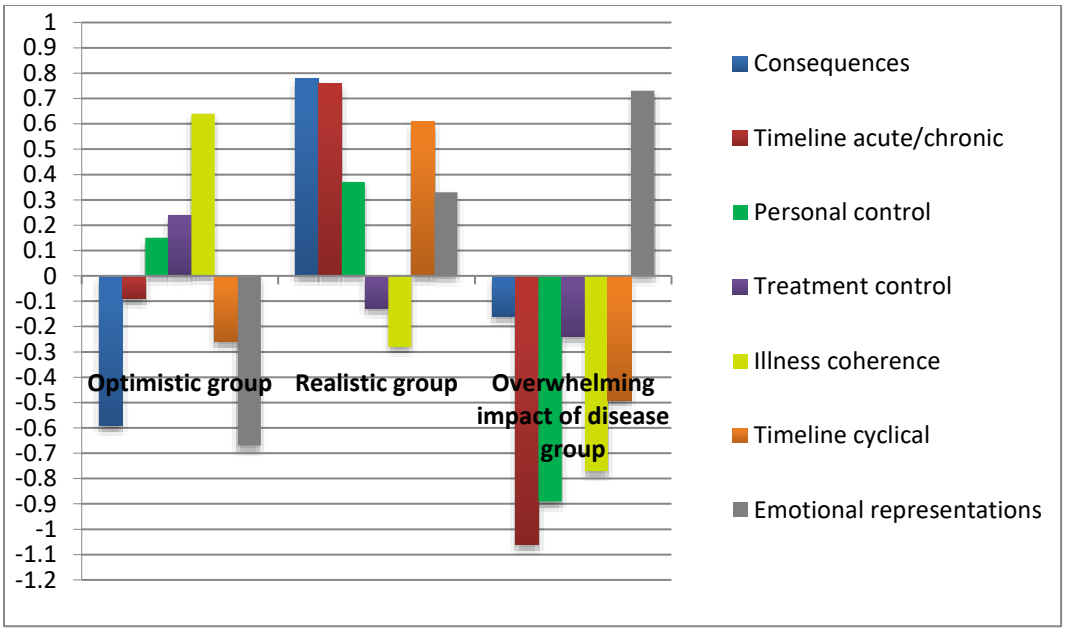
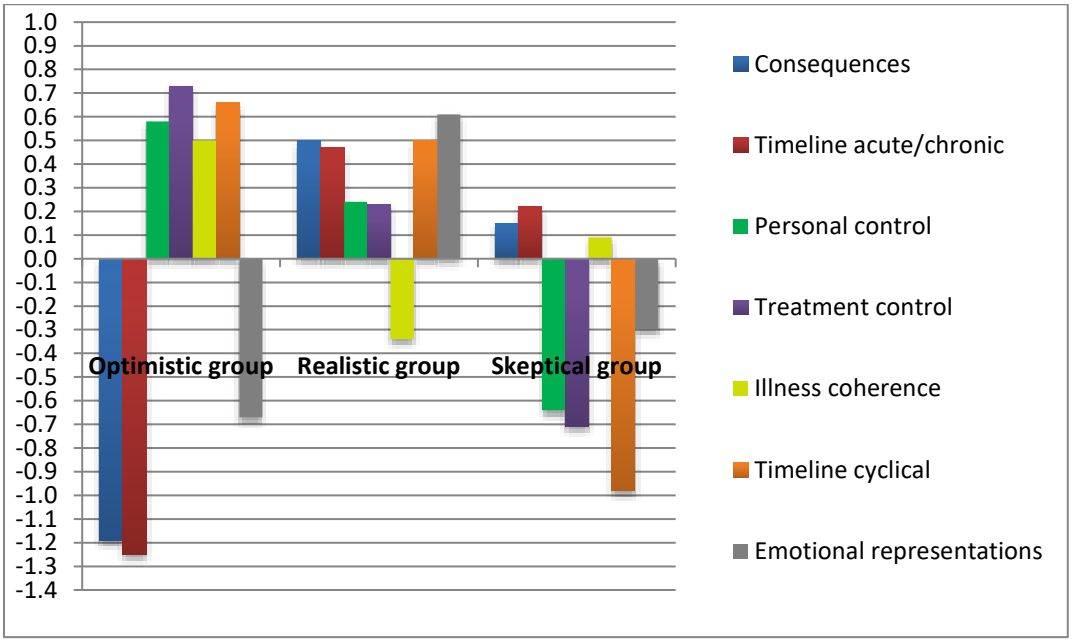


Figure 3: Mean z-scores of the illness perception dimensions per cluster for SSc



Characteristics of the physicians per cluster

The descriptive analysis and distribution per specialism of the three obtained clusters are shown in Tables 2 and 3, respectively.

For SLE, in the 'optimistic' group, the proportion of men and women was almost equal, with a mean age of 39 years (range 27–35) and a mean of 9 years of working experience (range 0–35). In the 'realistic' group, 72% men, with a mean age of 48 years (range 32–66) and a mean working experience of 18 years (range 0–46) were detected. In the 'overwhelming impact of disease' group, 64% male physicians with a mean age of 42 years (range 27–59) and a mean of 11 years of working experience (range 0–30) were present. Three of the five ophthalmologists were present in the 'optimistic' group. The 'realistic' group comprises three of the five rheumatologists and the distribution of the dermatologists and internists was the largest (3/5) in the 'overwhelming impact of disease' group.

For SSc, in the 'optimistic' group, 64% of the physicians were male with a mean age of 40 years (range 27–55) and a mean of 10 years of working experience. In the 'realistic' group, 76% were men with a mean age of 44 years (range 27–66) and a mean working experience of 14 years (range 0–46). In the 'skeptical' group, 56% women with a mean age of 44 years (range 28–65) and a mean working experience of 13 years were present (range 0–35). The distribution per specialism was as follows: the 'optimistic' group consisted of the two pulmonologists but none of the rheumatologists. The 'realistic' group comprised three of the five rheumatologists, dermatologists and gastroenterologists. Finally, 6 of the 11 nephrologists were present in the 'skeptical' group.

Table 2: Descriptive analysis and distribution per specialism of the 3 obtained clusters for SLE

	Cluster 1 Optimistic group	**Cluster 2 Realistic group**	**Cluster 3 Overwhelmed by disease group**
N	21	18	11
Age, mean (range)	39 (27-65)	48 (32-66)	42 (27-59)
Females (%)	48	28	36
Years' experience, mean (range)	9 (0-35)	18 (0-46)	11 (0-30)
Frequency of patient contact*	4 (1-6)	4 (1-6)	4 (1-6)
Dermatologists (n=5)	1	1	3
Ophthalmologists (n=7)	5	2	0
Gynecologists (n=5)	3	2	0
Rheumatologists (n=5)	2	3	0
Internists (n=6)	1	2	3
Cardiologists (n=4)	2	2	0
Pulmonologist (n=2)	1	1	0
Nephrologists (n=11)	4	3	4
Gastroenterologist (n=5)	2	2	1

*1= once a week; 2= several times a week; 3= once a month; 4= several times a month; 5= once a year; 6= several times a year

Table 3: Descriptive analysis and distribution per specialism of the 3 obtained clusters for SSc

	Cluster 1 Optimistic group	**Cluster 2 Realistic group**	**Cluster 3 Skeptical group**
N	11	21	18
Age, mean (range)	40 (27-55)	44 (27-66)	44 (28-65)
Females (%)	36	24	56
Years' experience, mean (range)	10 (0-25)	14 (0-46)	13 (0-35)
Frequency of patient contact*	4 (1-6)	4 (1-6)	3 (1-6)
Dermatologists (n=5)	0	3	2
Ophthalmologists (n=7)	3	3	1
Gynecologists (n=5)	2	2	1
Rheumatologists (n=5)	0	3	2
Internists (n=6)	1	2	3
Cardiologists (n=4)	1	2	1
Pulmonologist (n=2)	2	0	0
Nephrologists (n=11)	2	3	6
Gastroenterologist (n=5)	0	3	2

*1= once a week; 2= several times a week; 3= once a month; 4= several times a month; 5= once a year; 6= several times a year

4. Discussion

The aim of this study was to explore differences in physicians based on their perceptions of patients with SLE and SSc, using patient vignettes as a novel technique. We observed a large variability in illness perceptions among physicians from different disciplines and within physicians of the same discipline. Differences in illness perceptions of physicians were not based on the frequency of patient contact.

Closer inspection of the clusters based on the SLE vignettes revealed that physicians who belong to cluster 1 are more optimistic about the disease than the physicians in the two other clusters. A possible explanation for this is that they underestimate the severity and burden of this illness and are less familiar with SLE compared to their colleagues. They have the lowest years of working experience and are younger in comparison with the two other clusters. The same is applicable for the SSc vignettes.

Most of the rheumatologists are situated in the 'realistic' group for both SLE and SSc vignettes. For the SSc vignettes, the 'realistic' group had the best understanding of SSc, with the most years' of experience and was characterized by the least female physicians in comparison with the other clusters. We suppose that the 'realistic' group has another view in comparison with the other clusters because of more clinical experience and focus on psychosocial aspects. For the SLE vignettes, cluster 3 was called the 'overwhelming impact of disease' group because we assumed that the perceptions of these physicians are more driven by their perception that SLE has an overwhelming emotional impact on the patient. The latter cluster had some comparable perceptions with the perceptions of last-year medical students in the recent study of Nowicka-Sauer et al. [22]. In this study, doctors-to-be perceived SLE as being less controllable, more burdensome, having more consequences, more symptoms and more emotional impact. Cluster 3 of the SSc vignettes was called the 'skeptical' group because these physicians seemed somewhat doubtful or uncertain of the impact of the disease on the patient in comparison with the other two groups of physicians.

This study is one of the few to investigate the cognitive basis of physicians' perceptions of patients with rheumatologic conditions. Although evidence from all pathologies is limited, and the use of different methodologies or analysis techniques makes it difficult to compare study results, it is increasingly recognized that there are discrepancies in illness perceptions between medical staff from different disciplines. For instance, compared to oncology nurses, radiation therapists, overestimated the impact of treatment for, and perceived duration of, breast cancer [23]. In the study of Dickman et al.[24], doctors perceived irritable bowel syndrome and inflammatory bowel disease as more chronic, having less severe consequences and being less understandable for the patients than the nurses did. Moreover, staff working in a neuroscience ward have been shown to have a greater understanding of epilepsy and psychogenic nonepileptic seizures in comparison to emergency care

staff. The latter staff perceived psychogenic nonepileptic seizures as less chronic in comparison with the neuroscience staff[25].

There are a number of strengths and limitations of this study which should be considered. First, our use of case-based study research implies that generalizability may be limited. However, we chose to use vignettes as a tool for measuring illness perceptions, because previous evidence has shown that certain concepts such as perception, health or freedom are sometimes defined more clearly with reference to real-life patient examples [26]. Moreover, it would not have been possible to explore the views of different physicians who care for the same patient in a real-world setting because it is not realistic to investigate this for each individual patient.

Another limitation is that the sample was limited to physicians and that other healthcare professionals such as specialist nurses, physical therapists, social workers, psychologists, etc., were not included in the study. That being said, the participating physicians are a representative sample of physicians who were employed in the largest university hospital and two of the largest general hospitals in Belgium and likely to be actively treating patients such as those described in the vignettes. That said, some physicians had an infrequent exposure to these patient populations. There was no problem of selection bias because the sample of physicians, who have knowledge and expertise in these patient populations, was representative for Belgium. Belgium is a small country and in the hospitals that we contacted there were not many doctors with expertise in SLE or SSc. Second, we choose to use paper surveys instead of email surveys and used a personalized covering letter in order to enhance participation. Reminders were sent via email and the rheumatologists from the two general hospitals were also asked to stimulate participation.

The existence of these different physician profiles has consequences for daily clinical practice because when physicians with different beliefs about illnesses are responsible for giving information about the course, consequences and controllability of these diseases to patients discrepancies or confusions may arise. If physicians give distinct, wrong or insufficient information, based on their own perception, the communication between the patient and the doctor may be impacted, leading to mistrust [27]. Moreover, discrepancies between medical professionals' perceptions and their attitudes and patients' attitudes may affect patients' subsequent health behavior [28].

This raises the question about who is the best to coordinate these pathologies and how patient care should be managed and planned posing of course a fundamental challenge in organizing care by several physicians for these entities. Evidence about physicians' perceptions concerning the organization of care for patients with SLE and SSc is scarce. Only one viewpoint article [29] described a fictional debate between a dermatologist and rheumatologist about the management of SLE patients with a discrepancy between these two physicians on who can handle lupus patients best. In Belgium and other European countries, a certification of specialization in SSc or SLE does not exist.

This means that evaluating perceptions only in doctors specialized in SSc and SLE does not make sense. Nevertheless, we assume that a solution for the evaluation and treatment of complex systemic diseases such as SLE and SSc would be the organization of centers that can function as reference or expert centers and where integrated care is developed and guided.

In **conclusion**, our study has shown that physicians from a number of disciplines hold different perceptions on the consequences, controllability, time course, and emotional impact of these two rheumatic diseases. This highlights the need for healthcare providers to consider how best to manage and plan, not only patient treatment, but also patient communication. Although it is clear that careful consideration should be given to the consequences for organizing future care in these multisystem diseases to enhance the quality needs, we also propose that further research should focus on differences in perceptions between different health care professionals such as specialist nurses, psychologists, etc., to better understand factors that influence differences in perceptions and their consequences for patient care.

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Appendix 1: Description of the four patient vignettes

-The **first vignette** describes a 28-year old Systemic Lupus Erythematosus (SLE) patient with a positive auto-antibody profile, proteinuria and lupus nephritis. She develops an epileptic attack and medication is started by her treating physician for the epileptic attack and lupus nephritis. Hereafter, her condition stabilizes. The patient herself wants a dose reduction of the anti-epileptic medication but is also afraid for a new epileptic attack after the dose reduction.

-The **second vignette** describes a 38-year old man who develops diffuse Systemic Sclerosis (SSc) with organ involvement more specifically the heart, lungs and muscles are involved. He has also a more progressive skin fibrosis due to his condition. Medication to stop disease progression is started but his physical condition deteriorates. The organ involvement spreads with chronic diarrhea and erectile dysfunction as a result. After intravenous infusions of a biological medication, disease activity stabilizes and he can fulfill his daily activities. Currently, he works part-time in an administrative function. His functionality and coping with his chronic condition improved.

-The **third vignette** is about a 35-year old women, diagnosed with SLE, and who develops 9 years after diagnosis, lupus nephritis. Medication has been started but the renal function deteriorates. This patient has a relationship and an 8-year old son. She works fulltime as a shop assistant. At the last consultation she had no complaints due to her disease except some fatigue. Her medication was switched because of a pregnancy wish.

-The **fourth vignette** is concerning a 42-year old patient diagnosed with diffuse SSc and interstitial lung disease. A year after diagnosis, she complains of dyspnea, coughing and itching. The coughing is very disturbing because of social isolation. Two years after the diagnosis of SSc her lung function deteriorates. After several intravenous infusions of a biological medicine, her disease activity does not improve and her treating physicians decide to conduct autologous stem cell transplantation. Hereafter, her quality of life improves but she finds it difficult to cope with the uncertainty of her future and the unpredictability of her disease. At this moment, she has no relationship but gets most of the emotional social support from her brothers and friends.

5

ILLNESS REPRESENTATIONS OF SYSTEMIC LUPUS ERYTHEMATOSUS AND SYSTEMIC SCLEROSIS: A COMPARISON OF PATIENTS, THEIR RHEUMATOLOGISTS AND THEIR GENERAL PRACTITIONERS

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Abstract

Objective: Discrepancies in illness representations between patients and physicians result in treatment difficulties, decreased well-being of patients, but also to misunderstandings and disrupted communication. Hence, the objective of this study was to compare illness perceptions of individual patients with systemic lupus erythematosus (SLE) and systemic sclerosis (SSc), their rheumatologists and their GPs and explore potential differences.

Methods: This study has a cross-sectional design. Patients with SLE and SSc, who were followed at the rheumatology department of the University Hospitals Leuven (Belgium), completed the revised Illness Perception Questionnaire (IPQ-R) which measures patients' perceptions of their condition and captures 9 dimensions. Physicians completed the Revised Illness Perception Questionnaire for Healthcare Professionals (IPQ-R HP) which consists of 7 dimensions and measures perceptions of the healthcare professional regarding the disease of their patients. Intraclass correlation was performed to examine relationships between pairs of respondents; Cohen's d for estimating the magnitude of the difference.

Results: Questionnaires were sent to 284 patients of whom 241 (113 SSc and 128 SLE patients) were included. Five rheumatologists and 160 GPs participated. For both diseases, positive correlations were found for 'consequences', 'illness coherence' and 'emotional representations' between patients, rheumatologists and GPs. GPs scored higher on the 'consequences' of these diseases for the patient (d=0.71 for SLE; d=0.80 for SSc). Differences between rheumatologists and GPs were small for SSc and moderate to large for 'consequences' (d=0.56) and 'timeline acute/chronic' (d=0.95) in SLE with higher scores for GPs.

Conclusions: For both diseases and between the 3 groups significant correlations are detected for the dimensions 'consequences', 'illness coherence' and 'emotional representations'. Differences between rheumatologists and GPs were mainly detected in the case of SLE patients. This can have implications for the collaboration between these two groups of physicians in daily clinical practice.

1. Introduction

Systemic lupus erythematosus (SLE) and systemic sclerosis (SSc) are severe and complex chronic auto-immune diseases characterized by multiple organ involvement, a heterogeneous presentation and an unpredictable disease course often leading to important morbidity and mortality.[1,2] Both diseases can have an important impact on patients' quality of life and the ability to carry out activities at home or at work due to pain, decreased physical functioning, fatigue and dyspnea. [3–6] These progressive or recurrent symptoms might influence the perceptions patients have about their condition.

Ideas about illness are an essential part of the self-regulation model, which proposes that behavior in relation to illness depends on an individual's perception or representation of his/her condition. In this model, Howard Leventhal and colleagues (1984)[7] postulated that illness representations consist of five elements: identity (symptoms), cause, consequences (effects on life), time line (duration), and controllability or cure of the condition. Studies in patients with SLE have demonstrated that illness perceptions are related to outcomes such as changes in psychological well-being over time [8], sexual (dys)functioning[9], (non-)adherence to therapy [10] and in SSc with physical and mental health but not with disease related characteristics. [11,12]

Most studies only focus on patients' illness perceptions and their association with clinical or patient-related outcomes[13] but the extent to which healthcare providers' ideas about the consequences of a chronic disease in specific patients match with those expressed by individual patients, is unknown. This could be important because the management of SLE and SSc requires a therapeutic relationship between patients and providers over years, which makes adequate health care a joint responsibility of both providers and patients. Moreover, it is possible that because of this relationship for years, physicians' perceptions can be influenced by patients and vice versa.

In one of the first studies [14] describing beliefs about arthritis in patients and physicians (most of them were rheumatologists), differences were detected about what physicians think patients believe and what patients actually believe about causes of arthritis or what helps in arthritis. In a study about epilepsy and seizure disorders, there were differences between the illness perceptions of patients and their doctors, especially about the controllability of the condition, which could represent barriers to successful clinical management[15]. In another study about breast cancer[16], medical professionals' perceptions of the consequences of treatment and duration of cancer did not match patients' beliefs: oncology nurses underestimated, whereas radiation therapists overestimated the impact of treatment and perceived duration of the disease. A study in osteoarthritis and diabetes showed that incongruence in patients and general practitioners' perceptions regarding stressors accompanying chronic disease is larger in diseases with a less clear treatment policy and may influence healthcare use and physical and mental functioning [17].

So, detecting discrepancies between healthcare providers and patients is of utmost importance because these may lead to problems in treatment, decreased well-being of patients [18], but also to misunderstandings and disrupted communication [17,19,20]. Differences in illness perceptions between patients with systemic auto-immune diseases, rheumatologists and general practitioners (GPs) are likely to be relevant because the knowledge and disease-related experience of these 3 groups is different. This is the first study to investigate these differences and to attempt a direct comparison between physicians and particular patients with SLE and SSc they care for. Hence, the aim of this study was to investigate similarities and differences in illness perceptions of individual patients with SLE and SSc with that of their rheumatologists and GPs.

2. Methods

Design

The present study has a cross-sectional design. However, it was part of a larger longitudinal project in patients with SLE and SSc. This study has been registered in clinicaltrials.gov with IDnr. NCT02655640. The data for the current evaluation where those collected at baseline between November 2015 and February 2016.

Study population

The study population consisted of patients, rheumatologists and GPs. A total of 284 patients with SLE and SSc, who were in follow-up at the University Hospitals Leuven (Belgium) and fulfilled the inclusion criteria, were approached and invited for participation. The inclusion criteria were as follows: the patient's medical and cognitive condition allows him/her to complete questionnaires; the patient has no severe psychiatric problems; the patient is proficient in Dutch and able to complete the questionnaires in Dutch. The five rheumatologists who were asked for participation worked at the systemic diseases care program at the rheumatology department of the University Hospitals Leuven. In addition to the rheumatologists, also the patients' GPs were asked to participate.

Procedure

Patients received a letter with information about the goal of the study, a questionnaire pack and an informed consent form together with a pre-stamped envelope. Patients were asked to complete the informed consent and the questionnaire pack and return it within 2 weeks.

After the patients gave informed consent, the treating rheumatologist and GP of each patient were approached. The physicians were asked to fill out an illness perception questionnaire developed for

healthcare professionals after completing an informed consent form. While completing the questionnaire, they had to rely on the most recent medical and psychosocial situation of the patient. The physicians were requested to complete the questionnaires as soon as possible after the patient consulted them. A maximum interval of 6 months between the consultation of the patient and completion of the questionnaire by the physician was allowed. Additionally, for both patients and physicians 3 reminders were sent after 3 weeks, 5 weeks and 7 weeks. After 9 weeks, patients were contacted by telephone if they were persistent non- responders.

Measures

Demographic characteristics

Age, gender, educational level, employment status, social status and living situation were collected from SLE and SSc patients (see Table 1).

Clinical data

Disease duration and disease activity were measured. In SLE patients, disease activity was assessed by the Systemic Lupus Erythematosus Disease Activity Index (SLEDAI) score with the Safety of Estrogens in Lupus Erythematosus National Assessment (SELENA) modification (i.e. SELENA-SLEDAI).[21] The SLEDAI is a valid and reliable index, which measures disease activity within the last 10 days [22] including 24 weighted objective clinical and laboratory variables. Disease activity can range from 0 to 105. The following activity categories have been defined on the basis of SLEDAI scores: no activity (SLEDAI=0), mild activity (SLEDAI=1–5), moderate activity (SLEDAI=6–10), high activity (SLEDAI=11–19), very high activity (SLEDAI≥20)[23].

In SSc patients, the SSc disease activity index (2003)[24] was used for measuring disease activity. This is a preliminary validated index to assess disease activity in SSc which consists of clinical and laboratory measures of disease activity as well as measures of disease activity being assessed by the patient only. It captures 10 weighted measures and the scores have a range from 0-10. SSc is considered to be active if the disease activity is ≥ 3.

Illness perceptions of patients and physicians

Patients completed the Dutch version[25] of the revised Illness Perception Questionnaire (IPQ-R) developed by Moss-Morris and colleagues.[26] This questionnaire measures perceptions of patients regarding their disease and consists of 9 dimensions or subscales: an illness identity dimension, 7 illness perception subscales and a causal attributions dimension. It has demonstrated good reliability and

validity across several illness groups and is one of the most widely applied instruments for assessing perceptions about illness.[27]

For the purpose of this study, we only used the 7 illness perception subscales (38 items) that include views about how long the disease will last (timeline acute/chronic); the recurrent nature of the condition (timeline cyclical); perceived consequences of the condition; perceptions of personal control and treatment control; patient's overall illness comprehension (illness coherence); and emotional representations. The items for all subscales are rated by the patient on a 5-point Likert scale from 1 ('strongly disagree') to 5 ('strongly agree'). Scores were calculated as the sum of the items per scale (as in the original publication).

The physicians completed the Revised Illness Perception Questionnaire for Healthcare Professionals (IPQ-R HP)[28]. The IPQ-R HP [28] is an adapted version of the IPQ-R[26] which is devised to be completed by healthcare professionals (HP). It is a 38-item questionnaire rated on a 5-point Likert scale from 1 ('strongly disagree') to 5 ('strongly agree') and consists of the same 7 dimensions as described above. The key characteristic of the IPQ-R HP is that healthcare professionals are asked to indicate what they think that the perceptions of the particular patient are. Sample items read as: "The illness of my patient has major consequences on his/her life; "The illness of my patient will last for a long time"; or "The symptoms of the condition of my patient are puzzling to him/her".

Statistical analysis

Sociodemographic variables, clinical variables and self-reported data from the questionnaires were summarized using descriptive statistics such as frequencies, means and standard deviations (SDs). To check the normality of the data the Kolmogorov-Smirnov test was used.

Intraclass correlation was performed to examine agreement between the 3 groups of respondents regarding the 7 illness perception dimensions: rheumatologists and their patients; GPs and their patients; rheumatologists and GPs. Cut-off values for the intraclass correlation are: trivial: <0.1; small: 0.1–0.29; moderate: 0.3–0.49; large: 0.5–0.69; very large: 0.7–0.89; almost perfect: >0.9[29].

A paired t-test was conducted to detect differences in these 7 dimensions between the 3 pairs of respondents as described above. The choice for a paired t-test was made on the basis of the distribution of the data and the link between patients and physicians; and rheumatologists and GPs. In addition, to appraise the magnitude of potential differences in perceptions between the groups, Cohen's d was calculated to estimate how large the difference was between the mean scores on the dimensions for each group. The cut-off values for Cohen's d are as follows: small= 0.20-0.50; medium= 0.50-0.80; large= ≥0.80 [30].

Analyses were carried out with Statistical Package for the Social Sciences, SPSS version 24.0 (SPSS, Inc., Chicago, IL, USA).

Ethical approval

Ethical approval was obtained from the Institutional Review Board of the University Hospitals Leuven (IDnr. B322201526067).

3. Results

Respondents

Questionnaires were sent to 284 patients who fulfilled the inclusion criteria, of which 241 participated (113 SSc patients and 128 SLE patients). The 5 rheumatologists and GPs of these patients were also asked for participation. The rheumatologists completed together a total of 229 questionnaires. A total of 240 GPs received questionnaires, 160 of which returned completed questionnaires (response rate= 66.7 %) (See Figure 1 for detailed information). The demographic and clinical information of the patients is shown in Table 1.

The group of the rheumatologists consisted of 2 women and 3 men. The group of the GPs consisted of 99 men and 61 women. The gender distribution in the group of the non-responders was comparable to that of the responders (60.0% vs 61.9%). No information on other characteristics was available.

Figure 1: Flow diagram of inclusion procedure patients and physicians

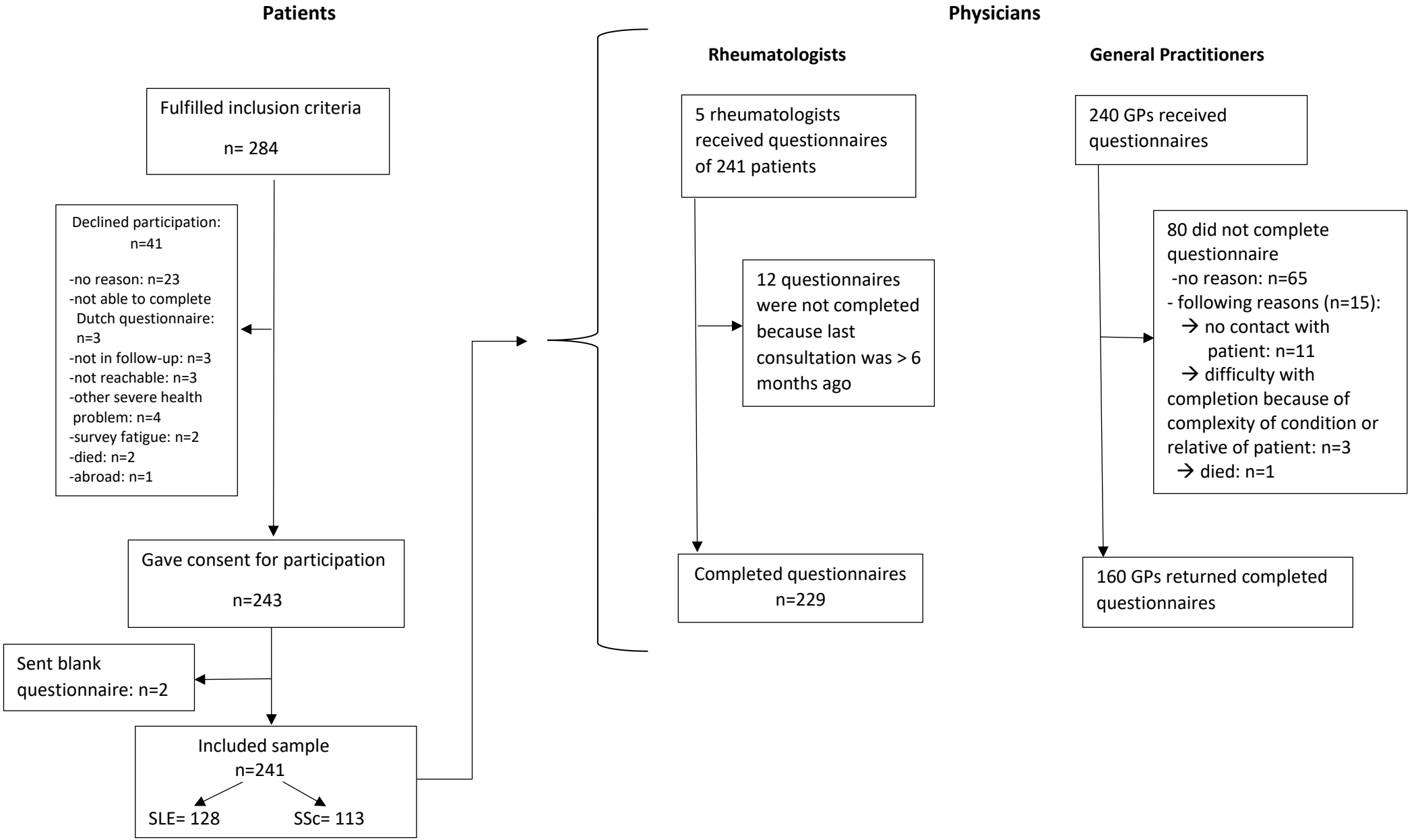

Table 1: Demographic and clinical characteristics of patients

	Systemic sclerosis (n=113)		**Systemic lupus erythematosus (n=128)**	
Gender				
Women (n-proportion)	n=76	(67.3%)	n=123	(96.1%)
Age in years (mean-SD)	60.17	(±10.82)	46.28	(±14.97)
Social status (n-proportion)				
Married	83	(73.5%)	78	(60.9%)
Cohabitation	3	(2.7%)	23	(18.0%)
Single	8	(7.1%)	11	(8.6%)
Divorced	12	(10.6%)	9	(7.0%)
Widow	7	(6.2%)	5	(3.9%)
Other	/	/	2	(1.6%)
Living situation (n-proportion)				
Living alone	20	(17.7%)	16	(12.5%)
Cohabitation with partner & kids	24	(21.2%)	46	(35.9%)
Cohabitation with partner	59	(52.2%)	53	(41.4%)
Cohabitation with kids	4	(3.5%)	6	(4.7%)
Cohabitation with friends	3	(2.7%)	7	(5.5%)
Other	2	(1.8%)	/	/
Education (n-proportion)				
Primary school	21	(18.6%)	10	(7.8%)
Secondary school	68	(60.2%)	58	(45.3%)
Bachelor degree	17	(15.0%)	41	(32.0%)
Master degree	7	(6.2%)	18	(14.1%)
Work status (n-proportion)				
Fulltime	14	(12.4%)	33	(25.8%)
Part-time because of illness	7	(6.2%)	13	(10.2%)
Part-time (personal choice)	3	(2.7%)	16	(12.5%)
Retired	49	(43.4%)	20	(15.6%)
Student	/	/	6	(4.7%)
Unemployed	2	(1.8%)	4	(3.1%)
Disablement benefit	18	(15.9%)	24	(18.8%)
Sickness benefit	8	(7.1%)	2	(1.6%)
Other	12	(10.6%)	10	(7.8%)
Disease duration in years (mean ± SD)	8.48	(±9.14)	13.90	(±9.31)
Disease activity (mean ± SD)	1.51	(±1.49)	3.40	(±3.27)

Comparisons between illness representations of SLE patients, their rheumatologists and their GPs (see Table 2)

Between patients and rheumatologists a large positive intraclass correlation was found for the perceived consequences of SLE. Moderate intraclass correlations were observed for controllability by treatment, illness coherence and the emotional impact of SLE. Looking at the Cohen's d, differences between patients and their rheumatologists were futile or small for all dimensions, except for timeline cyclical, on which the patients scored higher with a moderate difference.

Between patients and GPs, moderate positive correlations were found for the perceived consequences, illness coherence, and the emotional impact of SLE. A moderately large difference was found for consequences, on which GPs scored higher than their patients.

Between rheumatologists and GPs, moderate but statistically significant correlations were found for the perceived consequences, chronicity of the time course, illness coherence, and the emotional impact of SLE. For consequences, the difference in mean score was moderate, with higher scores in GPs than in rheumatologists, and for timeline acute/chronic, the difference was large.

Comparisons between illness representations of SSc patients, their rheumatologists and their GPs (see Table 3)

Also for SSc, moderate positive intraclass correlations were found between patients and rheumatologists on the consequences dimension, illness coherence and emotional representations. Patients scored lower in comparison with rheumatologists, with a moderate difference for the consequences dimension.

Between patients and GPs, moderate positive correlations were found for consequences and illness coherence and large intraclass correlations for personal control. Patients had lower mean scores on the perceived consequences with a large difference for consequences and moderate difference for timeline acute/chronic and illness coherence.

Between rheumatologists and GPs, large positive correlations were found for the perceived consequences, and moderate correlations for personal control, illness coherence and the emotional impact of SSc. A large difference was found for consequences, on which GPs scored higher than patients. For timeline acute/chronic and treatment control a moderate difference was found with also higher scores for GPs than patients. The detected differences between rheumatologists and GPs were small for all dimensions.

Table 2 : Comparison between SLE patients, their rheumatologists and their GPs based on 7 illness perception dimensions

	ICC Patients – Rheumatologists (n=115)	Patients (n=115) Mean (SD)	Rheumatologists (n=115) Mean (SD)	Cohen's d	ICC Patients-GPs (n=83)	Patients (n=83) Mean (SD)	GPs (n=83) Mean (SD)	Cohen's d	ICC Rheumatologists-GPs (n=79)	Rheumatologists (n=79) Mean (SD)	GPs (n=79) Mean (SD)	Cohen's d
Consequences	0.523‡	17.6 (5.3)	18.3 (5.6)	0.11	0.397‡	17.6 (5.1)	21.5 (4.3)	0.71	0.407†	18.2 (5.5)	21.4 (4.2)	0.56
Timeline acute/chronic	0.226	24.7 (3.8)	22.9 (3.2)	0.38	0.261	24.9 (3.6)	26.4 (3.3)	0.33	0.331†	22.7 (3.1)	26.3 (3.3)	0.95
Personal control	-0.076	18.6 (4.2)	17.3 (4.0)	0.22	-0.145	18.1 (4.4)	17.3 (4.3)	0.12	-0.108	17.3 (4.1)	17.4 (4.3)	0.02
Treatment control	0.360†	17.1 (2.8)	18.1 (1.9)	0.31	0.154	17.1 (2.9)	17.9 (2.4)	0.22	0.101	18.1 (1.7)	17.9 (2.3)	0.07
Illness coherence	0.388†	17.2 (4.2)	18.5 (4.3)	0.25	0.325*	16.9 (4.2)	18.9 (3.1)	0.46	0.415†	18.6 (4.4)	18.9 (3.0)	0.07
Timeline cyclical	0.158	14.7 (3.6)	12.1 (3.6)	0.54	0.152	15.2 (3.2)	13.6 (2.5)	0.42	0.102	11.9 (3.4)	13.6 (2.5)	0.40
Emotional representations	0.371†	16.7 (5.5)	16.1 (5.7)	0.08	0.440†	16.7 (5.5)	18.3 (4.1)	0.28	0.462‡	15.7 (5.6)	18.3 (4.1)	0.46

ICC: intraclass correlation coefficient; GP: general practitioner; *p≤0.05; †p≤0.01; ‡p≤0.001

Table 3: Comparison between SSc patients, their rheumatologists and their GPs based on 7 illness perception dimensions

	ICC Patients – Rheumatologists (n=111)	Patients (n=111) Mean (SD)	Rheumatologists (n=111) Mean (SD)	Cohen's d	ICC Patients - GPs (n=74)	Patients (n=75) Mean (SD)	GPs (n=75) Mean (SD)	Cohen's d	ICC Rheumatologists-GPs (n= 77)	Rheumatologists (n=77) Mean (SD)	GPs (n=77) Mean (SD)	Cohen's d
Consequences	0.339†	18.5 (4.8)	22.4 (5.7)	0.61	0.424‡	18.0 (4.7)	22.2 (4.6)	0.80	0.628‡	23.2 (5.3)	22.2 (4.6)	0.19
Timeline acute/chronic	0.078	24.9 (4.0)	25.9 (3.2)	0.20	-0.047	24.7 (4.1)	27.5 (2.7)	0.56	0.134	26.2 (3.2)	27.5 (2.6)	0.31
Personal control	0.166	17.1 (3.4)	17.9 (4.2)	0.16	0.575‡	16.9 (3.6)	16.1 (4.3)	0.18	0.371*	17.9 (4.6)	16.2 (4.3)	0.31
Treatment control	-0.040	14.9 (2.6)	16.6 (2.6)	0.44	0.168	15.0 (2.4)	16.7 (2.3)	0.55	0.108	16.4 (2.8)	16.7 (2.4)	0.09
Illness coherence	0.320*	15.8 (3.8)	16.1 (4.0)	0.07	0.439†	15.5 (4.1)	17.5 (3.4)	0.44	0.295*	15.2 (4.3)	17.5 (3.5)	0.46
Timeline cyclical	0.058	14.2 (3.2)	12.9 (2.9)	0.31	0.170	13.9 (3.1)	12.9 (2.2)	0.29	-0.191	12.6 (2.7)	12.8 (2.2)	0.04
Emotional representations	0.319*	18.1 (5.2)	19.7 (5.1)	0.24	0.040	18.4 (5.7)	18.0 (4.5)	0.05	0.313*	19.9 (5.1)	18.0 (4.5)	0.31

ICC: intraclass correlation coefficient; GP : general practitioner; *p≤0.05; †p≤0.01; ‡p≤0.001

4. Discussion

The aim of this paper was to explore commonalities and differences in perceptions between patients with SLE and SSc, their rheumatologists, and their GPs on the individual representations of their illness.

For both diseases, we found moderate to large correlations in the consequences, illness coherence and emotional representations dimension between patients, rheumatologists and GPs. The GPs scored higher on these dimensions in comparison with the patients were the difference was mostly small to moderate. They seemed to overestimate the consequences of these diseases for the patient, the understanding of these diseases by the patient and the emotional impact for SLE patients but not for SSc patients. Our results are in concordance with a number of studies in epilepsy [15,31,32] in which the neurologists score higher on the aforementioned dimensions than the patients. The rheumatologists scored also higher on these dimensions than the patients, but the differences were mostly small. Except in SLE, patients had the perception of a more recurrent time course than rheumatologists. This difference was moderate.

A closer look at comparisons between rheumatologists and GPs shows that for SLE, GPs have higher scores on consequences, illness coherence, timeline acute/chronic and emotional representations and for SSc, rheumatologists score higher than the GPs on these dimensions (except for illness coherence). This reflects that GPs consider SLE to be a much more severe condition than rheumatologists and patients do. This could be attributed to lack of knowledge or the fact that they rely on general information about SLE –for instance coming from textbooks, which tend to put too much emphasis on severe SLE manifestations- when scoring the questionnaire.

For SSc, moderate to large correlations were found for personal control in both patients and GPs and GPs and rheumatologists. The difference between these groups was statistically small but can have clinical implications. The fact that there is a difference can be related to the rarity of SSc, inter-patient differences, both in the type of controllable symptoms and the level of control[33] and that personal control is a construct of illness perceptions which is the most complex and multifactorial in relation to the other dimensions [34]. Another explanation is that GPs might have lack of knowledge or limited patient contact and therefore rely more on the information provided by the patient. This emphasizes the importance of the collaboration between GPs and rheumatologists for receiving and providing up-to-date disease-related information. In daily clinical practice, perceived control is based on knowledge about the disease -for instance provided by a healthcare professional- and also on the patients' personal experience. This dimension is important because of several reasons. The first reason is that personal control is one of the dimensions that predicts outcomes and is easily changeable with interventions. Much more than the other dimensions personal control can be altered by patient education sessions and self-management programs [35–37]. Dedicated educational programs with an

emphasis on the perceptions of patients, initiated by physicians, could reduce the knowledge deficit in the patients. Also, as the GP often remains the primary caregiver, it is crucial that the illness perceptions of both GP and rheumatologists are aligned. For this, communication modalities between these 2 groups of health professionals should be optimized.

Strengths of this study are that we had dyads of patients, rheumatologists and GPs and a high response rate. The number of GPs that participated was considerable. The study setting was a large university hospital in Belgium. The results of this study should be interpreted in light of some methodological limitations. A small number of GPs sent us a blank questionnaire with a note that they did not want to complete the IPQ-R HP because the patient visited them too long ago or not frequently so that they did not feel comfortable to judge in detail the patient's health situation. Another limitation is that the rheumatologists who completed the IPQ-R HP mentioned that completing the questionnaire was not always easy. Some dimensions such as for instance timeline acute/chronic, treatment control, personal control were mainly based on medical knowledge. Other dimensions such as the emotional representations and illness coherence, were completed much more subjectively by putting their selves in place of the patient. So, the set-up of the IPQ-R HP needs further exploration.

Another point for consideration is the external validity of the study results. This study has been conducted in a large single center which implies that the generalizability can be limited. Furthermore, the current study has a cross-sectional design which implies that it cannot establish the directionality of the associations between rheumatologists or GPs and patients. It is possible that the physicians' perception is influenced by the way a patient perceives and reports his condition, or vice versa. Other implications for further research are that the uncovered differences in illness perceptions between patients and medical staff can now be studied for their impact on outcomes. The current study provides insights in illness perception dimensions that are important for inclusion in future research about the influence of illness perceptions on outcomes such as for instance patient satisfaction, adherence, healthcare utilization, etc.

In **conclusion,** we can state that for both diseases and between patients, rheumatologists and GPs significant correlations in illness perceptions are detected for the dimensions consequences, illness coherence and emotional representations. For SLE, we found that GPs perceived the consequences of SLE as more severe than patients and rheumatologists do and that GPs also perceived SLE as more chronic than rheumatologists. GPs perceived SSc as more severe, chronic and controllable by treatment than patients. These differences can have implications for the communication and collaboration in daily clinical practice.

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6

PROSPECTIVE ASSOCIATIONS BETWEEN ILLNESS PERCEPTIONS AND HEALTH OUTCOMES IN PATIENTS WITH SYSTEMIC SCLEROSIS AND SYSTEMIC LUPUS ERYTHEMATOSUS: A CROSS-LAGGED ANALYSIS

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Abstract

Objectives: Literature states that illness perceptions of patients with systemic lupus erythematosus (SLE) and systemic sclerosis (SSc) are strongly associated with physical and psychological outcomes. This relation is most likely dynamic which means that outcomes can possibly predict perceptions of patients. This interplay is not fully investigated yet and clarity regarding the directions of associations is needed when designing interventions to alter illness perceptions. Therefore, this study aimed to investigate the prospective associations between illness perceptions and depressive symptoms, anxiety, perceived health status and disease activity in SLE and SSc patients.

Methods: Patients with SLE and SSc from a single-center university hospital participated in a longitudinal study spanning one year. Participants completed at both time points the revised Illness Perception Questionnaire; the HADS for measuring depressive symptoms and anxiety; the EQ-5D-5L for assessing perceived health status. Disease activity was also recorded. The directionality of the associations was investigated using cross-lagged path analysis controlling for age, gender and disease duration.

Results: A total of 128 SLE and 113 SSc patients with a mean age of 46.28 (±14.97) years and 60.17 (±10.82) years, respectively and disease duration of 13.90 (±9.31) years and 8.48 (±9.14) years, respectively participated. In SLE, reporting more depressive symptoms, more anxiety and worse perceived health status predicted a relative decrease in illness coherence one year later. More severe perceived consequences predicted a relative decrease in health status. The perception of a more chronic time course predicted an increase in depressive symptoms. In SSc, reporting more depressive symptoms and more anxiety predicted a relative decrease in illness coherence. A good perceived health status and less reporting of depressive symptoms predicted a relative decrease in perceived consequences.

Conclusions: Evidence was obtained for reciprocal pathways between health outcomes and illness perceptions although the predominant direction of effects was found to be from health outcomes to illness perceptions.

1. Introduction

Systemic lupus erythematosus (SLE) and systemic sclerosis (SSc) are characterized by multiple organ involvement, a heterogeneous presentation and an unpredictable disease course often leading to important morbidity and mortality [1,2]. Both diseases affect more women than men, with a sex ratio for SLE: 9:1 ratio and for SSc: 3:1 ratio. SSc is considered a rare disease (prevalence <1/5000) and has one of the highest mortality rates amongst all rheumatic diseases [3,4]. Beside the organ involvement, SLE and SSc patients might experience difficulties with personal care, household chores, work and leisure activities due to fatigue, dyspnea and impairments in physical functioning [5–8]. In addition to physical impairments, patients might experience psychological consequences such as depressive symptoms and anxiety. A recent meta-analysis stated that the prevalence estimates for SLE of major depression was 30% and major anxiety occurred in 40% of the patients [9]. Also, for SSc, this burden was high with 56% reporting major depression and 37% SSc patients having anxiety disorders [10] . Physical and psychological impairment might influence patients' illness perceptions, which are the mental constructions patients develop about their illness.

Researchers found that illness perceptions in SLE and SSc are associated with physical and mental functioning and other health outcomes such as sexual functioning, treatment adherence and depressive symptomatology [11–14] independently from disease-related characteristics or from the medical severity of the patients' condition. In the early 1980s, Leventhal and colleagues [15] conceptualized illness perceptions in the Common-Sense Model (CSM). The CSM shows that internal stimuli (e.g., symptom experience such as pain) and external stimuli (e.g., disease-related information from family or healthcare professionals) generate cognitive representations and emotional responses which guide the selection of coping procedures in order to eliminate and control potential or ongoing illness threats [16].

Research based on the CSM is mostly focused on illness perceptions predicting health outcomes but the relation is most likely dynamic which means that outcomes can possibly predict perceptions of patients. So, some of these health outcomes, such as anxiety and depression, perceived health or even disease activity as a more objective outcome, can potentially influence illness perceptions but this is not fully established or investigated [17]. Clarity regarding the direction of associations is needed when designing interventions to alter illness perceptions, as clinicians need to know how and where to intervene [18]. The available studies on illness perceptions and anxiety, depression, and perceived health status in patients with SLE and SSc are scarce and most of them are cross-sectional, except for one study [19] which describes correlations of illness perceptions with changes in psychological outcomes without information on the directionality of the associations. We assume- based on the CSM [15] that the predominant direction of effects goes from the illness

perception dimensions to subjective outcomes (i.e., depressive symptoms, anxiety, perceived health status) and also disease activity as an objective outcome. This hypothesis is based on previous literature in diabetes patients [20] which shows that illness perceptions precede the formation of depressive symptoms and stress over time. The aim of this study was to investigate the directionality of effects linking illness perceptions and health outcomes in SLE and SSc patients.

2. Methods

Design

The present study is a longitudinal observational cohort study of patients with SLE and SSc in which all variables of interest were measured at two time-points with an interval of 12 months. The data of time 1 were collected between November 2015 and February 2016 and the data for time 2 were collected between November 2016 and February 2017. This study has been registered in clinicaltrials.gov with ID nr. NCT02655640.

Study population

Patients were eligible for inclusion if their medical and cognitive condition allowed them to complete questionnaires; if they did not have severe psychiatric problems; if they were proficient in Dutch and were able to complete the questionnaires in Dutch. Overall, 284 patients with SLE and SSc who were in follow-up in our center fulfilled the inclusion criteria, and were therefore invited for participation.

Procedure

We sent at both time points a letter with information about the purpose of the study, a questionnaire pack and an informed consent form together with a pre stamped envelope to all eligible patients. They were asked to complete the questionnaires and the informed consent form and return it within two weeks. In case of non-response, reminders were sent after three weeks, five weeks and seven weeks. After nine weeks, patients were contacted by telephone if they were persistent non-responders. As an incentive, patients who completed the questionnaires at both time points received a voucher of 20 EUR.

Measures

Clinical data

Both disease duration and disease activity were measured. In SLE patients, disease activity was evaluated using the Systemic Lupus Erythematosus Disease Activity Index (SLEDAI) score with the Safety of Estrogens in Lupus Erythematosus National Assessment (SELENA) modification (i.e. SELENA-SLEDAI)[21]. The SLEDAI is a valid and reliable index that measures disease activity over the past 10 days [22]. It includes 24 weighted objective clinical and laboratory variables. The SLEDAI scores can range from 0 to 105 and allows to categorize patients into: no activity (SLEDAI=0), mild activity (SLEDAI=1–5), moderate activity (SLEDAI=6–10), high activity (SLEDAI=11–19), very high activity (SLEDAI≥20)[23].

In SSc patients, disease activity was measured using the SSc disease activity index (2003)[24]. This index consists of both self-reported data and clinical and laboratory measures of disease activity. It consists of 10 weighted measures and the scores can range from 0-10. An index score ≥ 3 reflects SSc that is active.

Illness perceptions

Patients' perceptions about their illness were measured with the Dutch version [25] of the revised Illness Perception Questionnaire (IPQ-R) developed by Moss-Morris and colleagues [26]. The IPQ-R is a self-report instrument which consists of 9 dimensions or subscales: an illness identity dimension, 7 illness perception subscales and a causal attributions dimension. It is a widely applied instrument across several disease groups and has demonstrated good reliability and validity [27]. The items for all subscales are rated by the patient on a 5-point Likert scale from 'strongly disagree' (1) to strongly agree (5). Scores were calculated as the sum of the items per scale. For the present study, we focused on 4 illness perception dimensions: the degree to which an illness was viewed as acute or chronic (timeline acute/chronic); the perceived seriousness of the condition (consequences); patient's perceptions regarding personal control they possessed over their illness; and patient's overall illness comprehension (illness coherence). The choice for these 4 dimensions was made because of reasons of parsimony regarding the applied statistical technique and was also based on previous findings from literature and correlational analyses.

Symptoms of anxiety and depression

Anxiety and depression were assessed with the Hospital Anxiety and Depression Scale (HADS) [28], a self-report questionnaire with seven items assessing anxiety and seven items assessing depressive

symptomatology. The HADS was chosen for use in this study because it is widely used, easily applied, avoids assessment of physical symptoms of depression, and has been validated in patients with rheumatic conditions and used in patients with SLE and SSc [9,10,29]. All items are scored on a 4-point scale from zero (not present) to three (considerable). The cut-off scores for the diagnosis of probable depression/presence of depressive symptoms is a score of ≥8 on the depression subscale and the cut-off scores for the diagnosis of probable anxiety was also a score of ≥8 on the anxiety subscale. The higher the score, the greater the degree of depressive symptoms and anxiety.

Perceived health status

Perceived health status was measured using the visual analogue scale of the EuroQol five-dimensions with 5 response levels (EQ-5D-5L) [30–32]. This standardized, self-report questionnaire consists of two parts: a health profile based on a descriptive system that defines health in terms of five dimensions and self-rated health. For assessing perceived health status, we used the second part of the EQ-5D-5L which measures the respondent's self-rated health on a visual analogue scale (EQ-5D VAS), with a score ranging from 0 (worst imaginable health status) to 100 (best imaginable health state) on the day of completion.

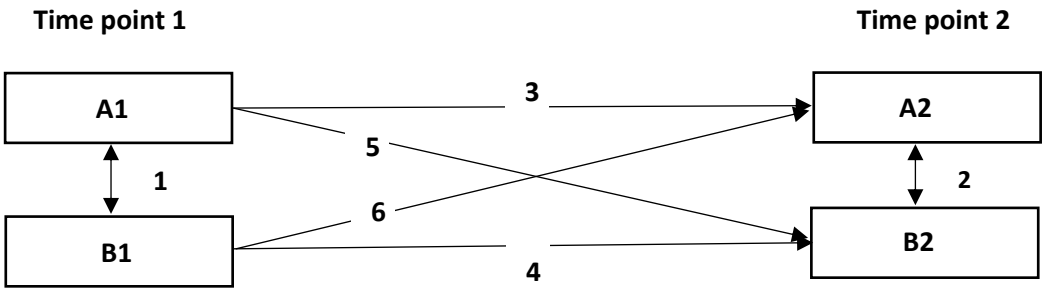
Statistical analysis

Cross-lagged analyses using structural equation modeling was conducted to examine prospective associations among illness perceptions and health outcomes (i.e. anxiety, depression, perceived health status and disease activity). A separate model was fitted for depressive symptoms, symptoms of anxiety, perceived health status, and disease activity. In all four models, all within-time associations, stability paths, and cross-lagged paths were estimated (except for the cross-lagged paths among the four illness perceptions). In addition, baseline age, gender, and illness duration were controlled for by estimating paths to each construct in the model. Only the significant paths with these control variables were retained in order to make the estimated cross-lagged model more parsimonious. Cross-lagged paths are an indication of the predominant direction of effects over time but should not be interpreted as definite proof of causation. In Figure 1, we described a cross-lagged model in which variables A and B are measured at two time points, resulting in three types of relations: within-time relations (1 and 2); autoregressive or stability relations (3 and 4); and cross-lagged relations (5 and 6). The estimated cross-lagged estimates can be interpreted as A1 predicting relative changes (i.e., relative increases or decreases) in B2 [33].

Maximum likelihood estimation with robust standard errors (MLR) was used to take into account the non-normality of the data. To assess model fit, the following fit indices were used: the

root mean square error of approximation (RMSEA, which should be <0.08); the comparative fit index (CFI, which should be >0.90) and the robust Satorra-Bentler scaled chi-square statistic (SBS χ^2) which should be as small as possible [34]. Data were analyzed with Mplus version 7. Missing data were dealt with using Full Information Maximum Likelihood (FIML).

Figure 1: Cross-lagged correlational model describing three types of relations adapted from Anderson & Kida (1982)



Ethical approval

All patients gave their consent for participation in the study and the Institutional Review Board of the University Hospitals Leuven provided ethics approval for this study (ID nr. B322201526067).

3. Results

Sample characteristics

Out of the 284 eligible patients, 241 participated (113 SSc patients and 128 SLE patients) at time 1 (response rate = 84.86%). These patients were asked to participate at time 2, of which 221 agreed (response rate= 91.70%). There was a drop-out rate of 8.3 % between times 1 and 2. Table 1 describes the demographic, clinical characteristics and health outcomes of the SLE and SSc patients at time 1.

Table 1: Overview patient characteristics at time point 1

	Systemic Sclerosis n=113	**Systemic Lupus Erythematosus n=128**
Gender		
Women (n-proportion)	76 (67.3%)	123 (96.1%)
Age in years (mean ± SD)	60.17 (±10.82)	46.28 (±14.97)
Social status (n-proportion)		
Married	83 (73.5%)	78 (60.9%)
Cohabitation	3 (2.7%)	23 (18.0%)
Single	8 (7.1%)	11 (8.6%)
Divorced	12 (10.6%)	9 (7.0%)
Widow	7 (6.2%)	5 (3.9%)
Other	/ /	2 (1.6%)
Living situation (n-proportion)		
Living alone	20 (17.7%)	16 (12.5%)
Cohabitation with partner & kids	24 (21.2%)	46 (35.9%)
Cohabitation with partner	59 (52.2%)	53 (41.4%)
Cohabitation with kids	4 (3.5%)	6 (4.7%)
Cohabitation with friends	3 (2.7%)	7 (5.5%)
Other	2 (1.8%)	/ /
Education (n-proportion)		
Primary school	21 (18.6%)	10 (7.8%)
Secondary school	68 (60.2%)	58 (45.3%)
Bachelor degree	17 (15.0%)	41 (32.0%)
Master degree	7 (6.2%)	18 (14.1%)
Work status (n-proportion)		
Fulltime	14 (12.4%)	33 (25.8%)
Part-time (because of illness)	7 (6.2%)	13 (10.2%)
Part-time (personal choice)	3 (2.7%)	16 (12.5%)
Retired	49 (43.4%)	20 (15.6%)
Student	/ /	6 (4.7%)
Unemployed	2 (1.8%)	4 (3.1%)
Disablement benefit	18 (15.9%)	24 (18.8%)
Sickness benefit	8 (7.1%)	2 (1.6%)
Other	12 (10.6%)	10 (7.8%)
Disease duration in years (mean ± SD)	8.48 (±9.14)	13.90 (±9.31)
Disease activity (mean ± SD)	1.51 (±1.49)	3.40 (±3.27)
Anxiety (mean ± SD)	6.77 (±3.54)	7.39 (±4.07)
Depression (mean ± SD)	5.59 (±3.71)	4.89 (±4.25)
Perceived health status (mean ± SD)	63.63 (±16.79)	68.32 (±15.64)

Cross-lagged analysis between illness representations of SLE patients and health outcomes

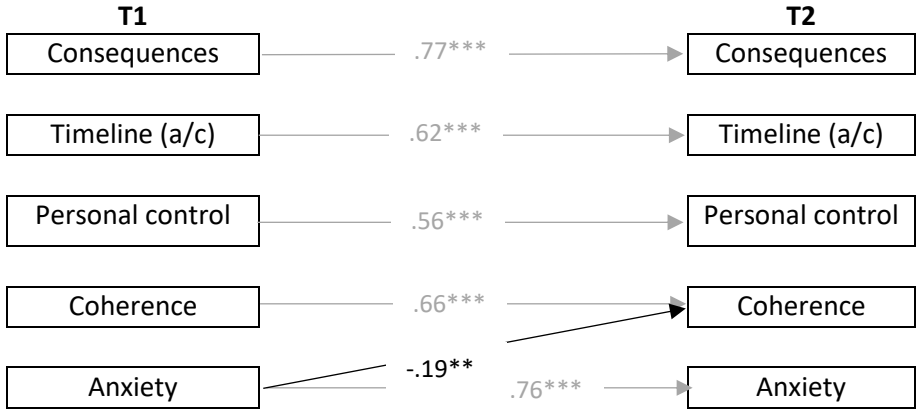
In Figure 2, all significant stability coefficients and cross-lagged paths for SLE are presented. The first cross-lagged model linking the four illness perceptions to anxiety (Fig. 2a) shows that high levels of anxiety predicted a relative decrease in illness coherence one year later. In the second cross-lagged model, described in Figure 2b, the presence of depressive symptoms predicted a relative decrease in illness coherence one year later. Furthermore, a chronic perception of the time course at time 1 predicted a relative increase in depressive symptoms at time 2. The third cross-lagged model (see Figure 2c) shows that high levels of perceived health status at time 1 predicted a relative increase in illness coherence at time 2 and that stronger perceptions of severe consequences predicted a relative decrease in perceived health status one year later. Finally, in the last model (see Figure 2d) no cross-lagged paths were found linking illness perceptions to disease activity. As shown in these figures, all models provided a good fit to the data.

Cross-lagged analysis between illness representations of SSc patients and health outcomes

In Figure 3, all significant stability coefficients and cross-lagged paths for SSc are presented. The first cross-lagged model, presented in Figure 3a, shows that high levels of anxiety predicted a relative decrease in illness coherence one year later. The second cross-lagged model in Figure 3b, shows that high levels of depressive symptoms at time 1 predicted a relative decrease in illness coherence and a relative increase in perceived consequences at time 2. Figure 3c, shows that high levels of perceived health status at time 1 predicted a relative decrease in perceived consequences one year later. In Figure 3d, only non-significant associations were present. As shown in these figures, all models provided a good fit to the data.

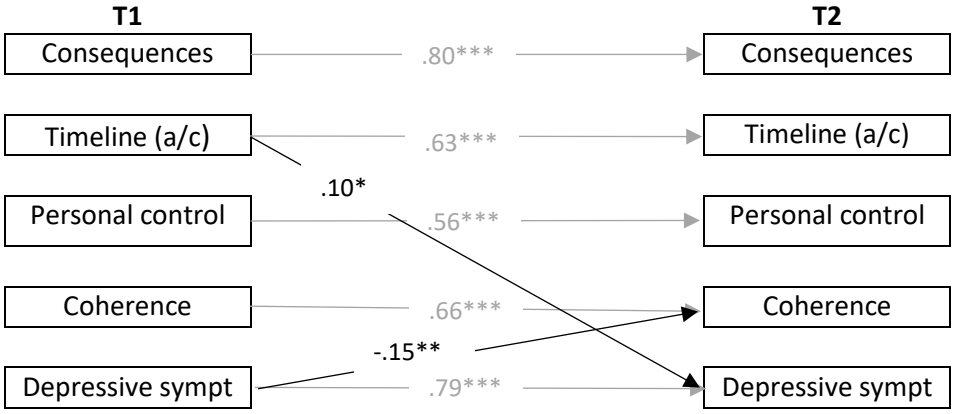
Figure 2: Overview of the cross-lagged path estimations between illness perceptions and health outcomes in SLE

Figure 2a: Cross-lagged model linking illness perceptions to anxiety



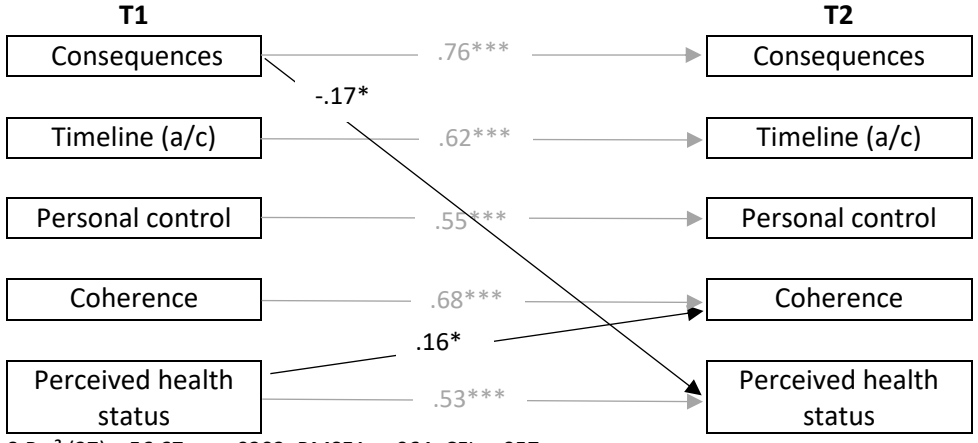
S-B χ^2 (36) = 52.22, p = .0393; RMSEA = .059; CFI = .966
**p < .01, ***p < .001

Figure 2b: Cross-lagged model linking illness perceptions to depressive sympt



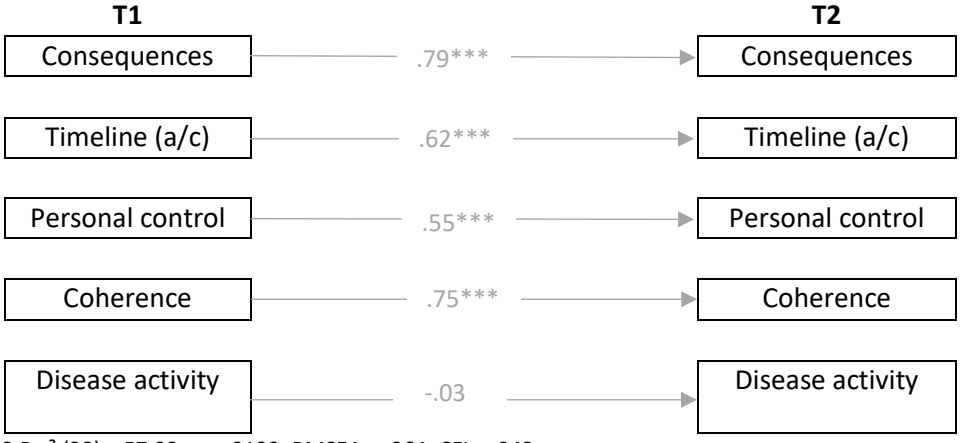
S-B χ^2 (37) = 55.77, p = .0245; RMSEA = .063; CFI = .962
*p < .05, **p < .01, ***p < .001

Figure 2c: Cross-lagged model linking illness perceptions to perceived health status



S-B χ^2 (37) = 56.67, p = .0203; RMSEA = .064; CFI = .957
*p < .05, ***p < .001

Figure 2d: Cross-lagged model linking illness perceptions to disease activity

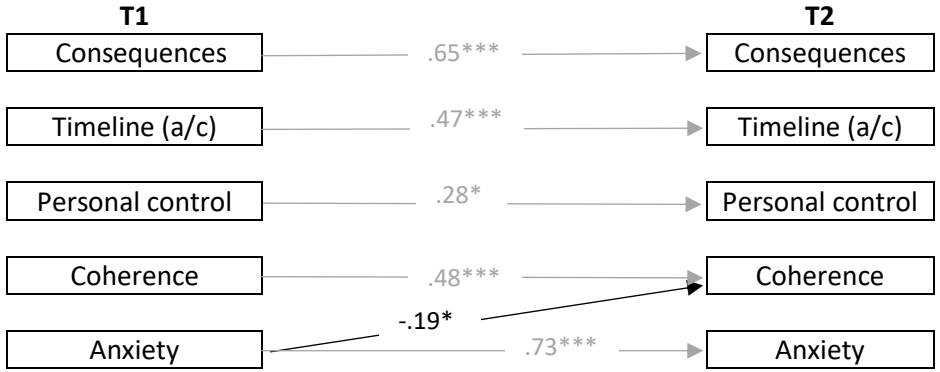


S-B χ^2 (38) = 57.99, p = .0199; RMSEA = .064; CFI = .942
***p < .001

Note: Within-time correlations and paths of the control variables (age, sex, and disease duration) are not presented for reasons of clarity. All path coefficients are standardized. Only significant stability and cross-lagged paths are demonstrated.

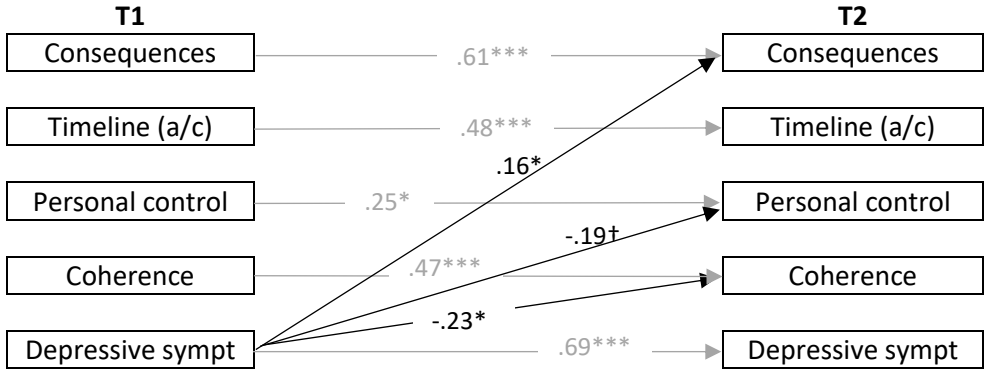
Figure 3: Overview of the cross-lagged path estimations between illness perceptions and health outcomes in SSc

Figure 3a: Cross-lagged model linking illness perceptions to anxiety



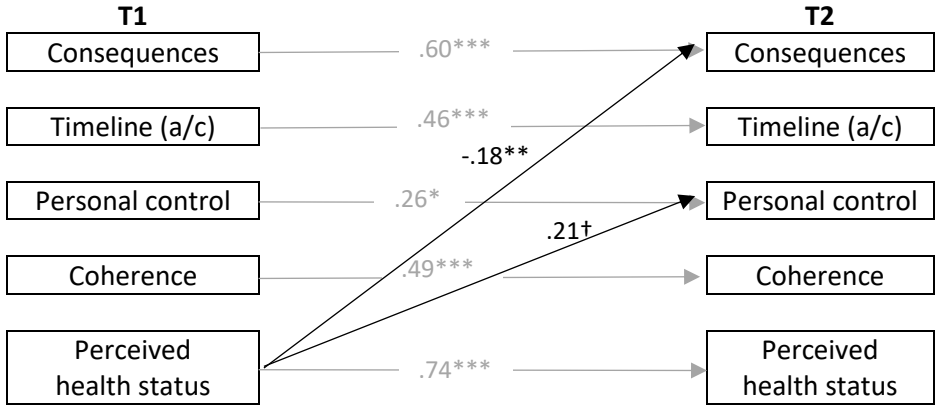
S-B $\chi^2(37) = 30.11$, p = .7817 ; RMSEA = 0; CFI = 1
*p < .05, ***p < .001

Figure 3b: Cross-lagged model linking illness perceptions to depressive symptoms



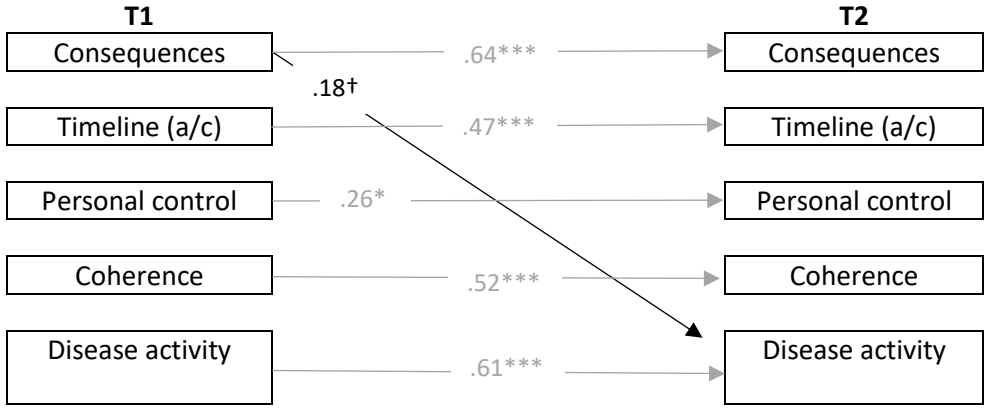
S-B $\chi^2(37) = 35.39$, p = .5447 ; RMSEA = 0; CFI = 1
†p < .10, *p < .05, ***p < .001

Figure 3c: Cross-lagged model linking illness perceptions to perceived health status



S-B $\chi^2(37) = 36.41$, p = .4964 ; RMSEA = 0; CFI = 1
†p < .10, *p < .05, **p < .01, ***p < .001

Figure 3d: Cross-lagged model linking illness perceptions to disease activity



S-B $\chi^2(37) = 38.78$, p = .3893 ; RMSEA = .021; CFI = .991
†p < .10, *p < .05, ***p < .001

Note: within-time correlations and paths of the control variables (age, sex, and disease duration) are not presented for reasons of clarity. All path coefficients are standardized. Only significant stability and cross-lagged paths are demonstrated.

4. Discussion

This study was the first to investigate prospective associations among illness perceptions and health outcomes in patients with SLE and SSc. For both diseases, we found that the predominant direction of effects goes in most cases from depressive feelings, anxiety and perceived health status to the illness perception dimensions.

For SLE patients, we found that high levels of anxiety, experiencing depressive feelings, and a low perceived health status predicted relative decreases in illness coherence one year later. No other illness perception dimensions were predicted by these outcomes. Illness coherence is the degree to which a person holds a coherent understanding of the illness; it is a metacognition that taps whether the illness "makes sense" to the patient [17]. Our results state that when patients report more depressive feelings or perceive their health status as worse, this may lead to a less coherent understanding of the disease. Broadbent and co-workers [20] mention that illness coherence can be seen as a summary of how the other perceptions fit together to provide a coherent model rather than being an independent illness perception per se. So, experiencing depressive symptoms may activate beliefs about the disease in general which implies that patients can become insecure about their condition. A closer look at the illness perception dimensions shows that perceiving the illness as more chronic predicts depressive symptomatology and that more perceived consequences predicted a poorer perceived health status in the case of SLE. Cross-sectional literature about illness perceptions in SLE and depression also found that the more chronic the perception of the disease, the more depressive symptoms. The finding that more severe consequences predicted worse perceived health status is in line with literature concerning other conditions [36] and also in line with literature stating that the consequences dimension is often a major factor in explaining or predicting outcomes [18]. Patients also report this in daily practice. Patients who experience consequences on social or financial level, for instance SSc patients who lost work because of their condition or SLE patients who experience fatigue, have a more negative view on their condition[19,35].

For SSc patients, we found that high levels of anxiety and depressive symptomatology predicted relative decreases in illness coherence and increase in perceived consequences and personal control, in the case of depressive symptomatology, one year later. A positively perceived health status also predicted less perceived consequences due to the disease which is comparable with what we found in SLE patients. Probably, the explanation for this is that in SSc the skin involvement and damage is more visible and debilitating in SSc patients which can explain why in SSc perceived health status precedes the formation of the consequences dimension[36].

A closer look at disease activity shows that in the SLE group disease activity levels were low and that no cross-lagged associations were found linking illness perceptions with disease activity. This

is in line with a SLE study [19] published 20 years ago and also with Hagger and Orbell (2003) [37] who state that illness perceptions are unrelated or weakly related to disease state. A possible reason is that disease activity is measured by the SLEDAI, an index which is solely based on more 'objective' biochemical and clinical characteristics and no 'subjective' patient reported characteristics or measurements. In SSc, the perceived consequences of the disease predicted disease activity at a marginally significant level.

We found that outcomes predominantly predicted illness perceptions and that perceived consequences and timeline acute/chronic predicted relative change in outcomes in SLE. Our findings stress the importance of holistic care and implies that healthcare professionals need to pay attention to their patients' perceptions, psychological wellbeing and perceived health status. Inaccurate perceptions can be tackled with specific counseling techniques, e.g. cognitive behavioral therapy. because Before drawing up further practical implications, further research in this area is needed.

Some limitations need to be taken into account when interpreting the study results. Kline (2005) [34] described that for structural equation modeling a sample of 5-10 respondents per variable is needed which was not feasible in the present study. We need to emphasize the relative rarity of these complex diseases and the exploratory nature of this study. Another limitation is the single-center setting which can reduce the generalizability of the findings to other settings.

Despite these limitations and suggestions for future research, the present study has also several strengths. Although this is a single-center study, the number of patients per disease group is appropriate because SSc and SLE have a rather low prevalence rate in Belgium and Europe compared to the US and Southern countries. Furthermore, these two diseases are sample cases of complex chronic diseases with an unpredictable course which makes extrapolation to other chronic conditions possible. Another strength is that we had a very good response rate at both time points. At time point 2, only 8.3 % of eligible patients did not participate. A possible reason for the high response rate are incentives and personal reminders in order to reduce non-response.

In **conclusion**, we can state that for SLE and SSc, anxiety, depressive feelings and perceived health status predict illness perceptions. Also for SLE, perceived consequences and the perception of a chronic disease course predicted perceived health status and depression, respectively. The finding that health outcomes precede illness perceptions shows that self-regulation is dynamic as described in the CSM.

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7

GENERAL DISCUSSION

Major progress in the understanding and treatment of severe complex diseases like multisystem auto-immune diseases has already been achieved or is awaited achievement in the near future but still witness a high unmet medical need. Classical innovative research is mainly focusing on somatic disease processes while aspects of illness perceptions and illness behavior also contribute importantly to outcome. Illness perceptions do matter since they have been reported in research concerning chronic diseases to be stronger associated with outcomes than purely disease related characteristics [1].

A closer look at the concept of illness perceptions, shows that when patients are confronted with a chronic condition, such as SLE and SSc, the type and quality of the available information provided to them, is crucial in order to give meaning to and manage their illness. Interpreting this information is the first step in the self-regulation of illness [2,3] and could be influenced by several factors which play an important role in establishing and shaping illness representations [4]. Since not all factors are already identified, it is of interest to have an overview of these and specifically the modifiable ones that could be adjusted by an intervention. One possible modifiable factor influencing patients' illness perceptions is the perception of healthcare professionals regarding the illness of a patient. Literature states that there are differences in illness perceptions between patients and their healthcare professionals [5]. Understanding the perceptions of various physicians concerning a particular patient they care for, could contribute to detect discrepancies with the patient's perception and to minimize or avoid eventual miscommunication or misconceptions.

Another point of attention is that the directionality of the associations between illness perceptions and health outcomes –such as anxiety, depression, mental and physical functioning- remains unclear in literature. On the one hand, publications state that health outcomes are correlates or determinants of illness perceptions, while on the other hand, others describe the importance of illness perceptions in influencing or predicting those health outcomes.

This PhD thesis identifies the modifiable correlates of illness perceptions and sheds light on the perceptions of physicians who are involved in the multidisciplinary care of patients with SLE and SSc. For measuring the perceptions of these physicians a preliminary validation of the illness perception questionnaire for healthcare professionals (IPQ-R HP), was conducted. Furthermore, we investigated if specific modifiable correlates of illness perceptions are to be considered as determinants or as outcomes of illness perceptions in patients with SLE and SSc.

In the next sections, reflections about the different chapters are summarized in terms of the value of this thesis to amplify the literature concerning illness perceptions, methodological considerations, implications for clinical practice, healthcare policy recommendations and horizons for further research.

1. Added value of this PhD study to the unknowns in literature

It is known that for developing effective interventions, researchers need to know how and where to intervene. In this regard, more research was needed to investigate the determinants or correlates of illness perceptions [6]. We provided an overview of the modifiable correlates of illness perceptions in Chapter 2 and found following factors: medication beliefs, information provision and satisfaction with received information, quality of care such as trust in the physician and perceived chronic care quality, illness-related factors and psychosocial factors. Such overview is important for HPs working in clinical practice because this added knowledge about the modifiable correlates of illness perceptions can structure patient education sessions and a HP's history taking. Working models for patient education [7] can be extended with this overview in order to tackle the formation of maladaptive illness perceptions.

Secondly, we conducted a preliminary validation of the IPQ-R HP (Chapter 3). The IPQ-R HP appears adequate and useful to assess the perception of HPs concerning the illness of an individual patient and produces -in this preliminary phase- reliable and valid output. Until now, researchers did not use a validated but a partially validated instrument for research purposes. In previous studies a modified version of the Brief-IPQ was used for measuring illness perceptions of medical students or nurses [8,9]. The preliminary version of the IPQ-R HP, validated in this thesis, can be utilized for research purposes but needs further psychometric exploration before application in daily clinical practice.

Thirdly, by measuring illness perceptions in rheumatologists, GPs and their patients using the IPQ-R HP (Chapter 5), we found small differences between rheumatologists' and GPs' perceptions in context of SSc and moderate to large differences for the dimensions 'consequences' and 'timeline acute/chronic' in context of SLE with higher scores for GPs in comparison with rheumatologists. Between patients, rheumatologists and GPs positive correlations were found for 'consequences', 'illness coherence' and 'emotional representations'. The uniqueness of this study was not only that we looked for commonalities and differences in illness perceptions in the aforementioned groups but that we had pairs of physicians and patients. The physicians were asked to complete the IPQ-R HP based on the knowledge and perceptions they have about their own patients, i.e. patients they know and treat in real life and not patient cases or diseases in general. This is in contrast to almost all previous illness perception research in HPs or students which is based on fictive patients or on a disease as such.

Fourthly, we answered an intriguing question, posed by researchers [2,6] experienced in illness perception research: are illness perceptions causally related to physical and psychological outcomes or vice versa? In Chapter 6, we conducted a cross-lagged analysis which shows the predominant direction of effects between illness perceptions and health outcomes such as anxiety symptoms,

depressive symptoms and perceived health status over time. We found that health outcomes at time point 1 predicted illness perceptions, mostly 'illness coherence' at time point 2 for both SLE and SSc patients and 'consequences' at time point 2 for SSc. Only the dimensions 'timeline acute/chronic' and 'consequences' in SLE predicted depressive symptoms and less perceived health status one year later, respectively.

Fiftly, we conducted the first longitudinal study in patients with SSc that investigates associations between illness perceptions and health outcomes. All other studies conducted in the area of illness perceptions in SSc have a cross-sectional design. For SLE, only one longitudinal study [10] in the format of a conference paper was available without the investigation of prospective associations between illness perceptions and health outcomes. In Chapter 6, we found that illness perceptions of patients with SLE and SSc are relatively stable over time. The fact that illness perceptions are stable over time is comparable with other studies in other diseases such as irritable bowel disease, diabetes or low back pain were the follow-up period ranged between 6 months and 2 years [11–13].

2. Reflections on the conducted studies: focus on participants and methods

Description of the patient population (at baseline)

The population of interest in this thesis comprised patients with two different diseases, i.e. patients with SLE and patients with SSc. This means that there was a certain variability in our cohort study. This variability was present at different levels. Firstly, we had within the group of SSc patients, two subgroups: the lcSSc subtype and dcSSc subtype. The difference between these subtypes lies in their severity; lcSSc has limited organ involvement, whereas dcSSc is generally perceived as a more severe disease with a lower life expectancy. We chose not to make a distinction between the subgroups of SSc in our studies because previous research in SSc, regarding the impact of illness perceptions on mental and physical functioning, concluded that the subtype of SSc does not contribute to differences in illness perceptions [14].

Furthermore, disease activity between these two illness groups was measured with two different measures as appropriate. For SLE patients, we used the SLEDAI and for SSc patients we used the SSc disease activity index. The difference between these two indices lies not only in their disease specificity but also in the composition of the parameters of the instruments. Concretely, the SLEDAI includes only objective parameters for measuring disease activity; the SSc disease activity index includes beside objective parameters also three subjective measures assessed by the patient only. These are: worsening of cutaneous involvement, worsening of vascular symptoms, and worsening of cardiopulmonary symptoms. Therefore, the SSc disease activity index might have correlations with illness perception measurements, which is not the case for the SLEDAI. Literature describes the lack of

correlations between disease activity measured by SLEDAI and subjective measurements of disease activity and also poor correlations between disease activity and quality of life in SLE patients [15,16]. Another important remark is that we were only able to measure disease activity in SSc patients in follow-up at the Belgian Systemic Sclerosis Cohort (BSSC). For the other SSc patients, not included in the BSSC, no data on disease activity was present because this is not a standard measurement in daily clinical practice. The same was applicable for disease activity in SLE patients. Also for this group no standardized measurement of disease activity in daily clinical practice is available. The SLEDAI was completed for the purpose of this study by the researchers and/or rheumatologists.

Another remark is that the study sample was relatively small but one has to be aware that SSc is a rare and SLE a less prevalent disease. This implies that a multi-center study could be the best way to have a larger sample size for the cross-lagged analysis conducted in Chapter 6 but this was currently not feasible.

Cross-lagged analysis: what and why?

A cross-lagged analysis is an analysis technique which investigates the predominant direction of effects over time. It is an indicator of temporal precedence and not a positive proof of causation [17]. In other words, it is a technique which gives more information about which variable precedes another variable. Within this analysis technique, one can control for the within-time relations, stability relations (i.e. between two time points) and also for background variables. The effect sizes of cross-lagged associations used in longitudinal research are much smaller than effect sizes in cross-sectional studies. The reason is that this analysis technique controls for stability relations in order to predict change in levels of the outcome over time which implies that it reduces the magnitude of the effect [18].

In our research, we were interested in this predominant direction of the associations between variables over time, because previous research in SLE and SSc, conducted in the area of illness perceptions, is of cross-sectional nature and assumes that the direction of the associations goes from illness perceptions to health outcomes. The performed longitudinal research in illness perceptions of patients with chronic conditions is generally focused on intervention research or on the capacity of (mal)adaptive illness perceptions to predict outcomes assuming that the direction of the relationship between illness perceptions and outcomes starts from illness perceptions. So, there is no clear answer on the question whether the illness perceptions precede the health outcomes or vice versa. In other words, in case of anxiety: do patients view their illness more negatively because they are anxious, or do they become anxious because they consider their illness so severe and feel they have no control over it?

The sample size needed for a cross-lagged analysis technique is between 5 to 10 respondents per variable [19]. We had data from 113 SSc patients and 128 SLE patients at baseline, satisfying the needed number of patients to calculate the cross-lagged correlations. Furthermore, this amount of data helped to form a basis for hypothesis building in a patient population where research was virtually not existing. In order to obtain a larger sample size, we could have studied these diseases as one systemic disease group. However, since SLE and SSc are different diseases with different phenotypes and according to the Common-Sense Model (CSM) which states that illness representations are generated and shaped by the experience of disease biology, we analyzed them separately. Another remark is that illness perceptions are not stand-alone constructs. This means that you cannot rule out that between time point 1 and time point 2 a stressful event occurs that can influence patients' perceptions. Moreover, such an event could not be controlled for because one does not know exactly when it happened. A possible solution for this is a diary study during 1 year where patients are asked to complete a daily diary in order to record thoughts and actions. The disadvantage of this research method is that it is an intensive and expensive design and that it might artificially increase ruminating about the disease and possibly induce inappropriate illness behavior.

The IPQ-R HP

The initial reason for developing the IPQ-R HP was in first instance the need for a questionnaire that measures illness perceptions of physicians and HPs. We adopted the IPQ-R as the basis for constructing the HP's questionnaire. Another reason was that there was a need for a questionnaire that resembled the IPQ-R, because in that way comparing HPs' responses and patients' responses was much more easy.

Why the IPQ-R and not the Brief-IPQ as a backbone for the IPQ-R HP?

We chose the IPQ-R, instead of the Brief-IPQ, for measuring illness perceptions in patients and as the core for developing the IPQ-R HP because of several reasons. The first reason was that the Brief-IPQ does not count all illness perception dimensions. For instance, the Brief-IPQ does not provide a detailed analysis of patients' identity beliefs and gives no information on cyclical timeline beliefs. For SLE and SSc patients, the recursive pattern and unpredictable disease course are disease specific and from that perspective also important. The second reason is that a single-item measure, such as the Brief-IPQ, is less reliable than a multi-item measure. Single items must capture the content of the dimension succinctly and internal reliability measurements are not possible [20]. The third reason is that Broadbent and colleagues [21] also mention that the IPQ-R in comparison to the Brief-IPQ may also be more sensitive to changes in illness perceptions due to the larger score range of the subscales. On the

other hand, a possible disadvantage of the IPQ-R is that it is longer and therefore more time consuming for HPs. However, HPs who completed the IPQ-R HP, did not specify this as a problem.

For patients, the completion of the IPQ-R and other self-report questionnaires was feasible judging by the high response rate at both time points. We offered them a voucher of 20 EUR after they completed the questionnaire pack at both time points. Probably, the incentive was large enough to motivate them for completing the questionnaires twice.

Impressions of rheumatologists concerning the structure of the IPQ-R HP

The five rheumatologists who completed the IPQ-R HP relied upon medical knowledge for the completion of some dimensions and on the patients' perspective for other dimensions. Rheumatologists stated that when there was a regular follow-up, such as in the BSSC, it was easier to complete the IPQ-R HP because they had more patient contact and therefore knew that particular patient better. Rheumatologists were surprised about the results in Chapter 5. The answers from rheumatologists on the questions differed less from patients than GPs' answers. However, rheumatologists did not expect this because they assumed that GPs knew their patients for a relatively longer period and are much more acquainted with them than rheumatologists are.

Rheumatologists also mentioned that completing the IPQ-R HP was not always easy. After the longitudinal study was conducted we went through all items of the IPQ-R HP and asked rheumatologists' thoughts about each item separately. They mentioned that some questions, for instance the items in Timeline acute/chronic, resembled each other or that there was some repetitiveness; or that some questions were unclear because they wondered if these items concerned the disease or symptom experience. Their completion of some dimensions such as for instance timeline acute/chronic, treatment control, personal control were mainly based on medical knowledge with sometimes the 'disease' in mind and not the particular patient. Other dimensions such as the emotional representations and illness coherence, were completed much more patient focused by putting themselves in place of the patient which was perceived as rather difficult. Some rheumatologists were also bothered by the fact that the IPQ-R HP was a generic questionnaire.

The advantage for rheumatologists of completing the IPQ-R HP was that they now had to overthink and deal with aspects they did not focus on previously or where they lacked knowledge. Rheumatologists also brainstormed about terminology of certain items such as the difference between 'fear' and 'anxiety' and reflected on questions regarding financial consequences of the disease for the patient because they were not aware of this. One rheumatologist mentioned that -in the case of dcSSc which is the most severe form of SSc- the consequences of the disease were not always communicated

to the patient. The reason was that the rheumatologist wanted to provide this information only when judged appropriate without worrying the patient too much.

We also wondered if rheumatologists were in agreement with each other on the scores of the IPQ-R HP items. We asked them to complete the IPQ-R HP based on four patient vignettes and found that the interrater agreement between the rheumatologists was low. This indicates that they perceived the same patient differently which can be attributed to the fact that their perceptions are subjective and that some of them did not work in centers where an integrated care program with regular patient case discussions was organized.

When should a healthcare professional complete the IPQ-R HP?

The IPQ-R HP should be preferably completed by the HP or physician at the moment when also the patient completes the IPQ-R. Since, illness perceptions are relatively stable, the IPQ-R should be completed at those time points when illness perceptions are prone to change. Leventhal and colleagues [22] described that illness representations are constantly being updated when new illness experience and knowledge regarding the illness are acquired. They stated this with an acute illness model in mind. Whereas for chronic diseases, which are relatively stable in time, aspects at disease start, or at diagnosis, would be more expected to induce change in perceptions [23]. Also, fluctuations in illness perceptions are detected during flares or acute exacerbations of the patient's condition [24]. So, illness perceptions will change when symptom experience changes [13], when there are changes in the clinical states of the patient or treatment [11].

In Chapter 3, we described that these questionnaires can be completed in an inpatient or outpatient setting. For instance, when patients are waiting in the waiting room. The doctor or other HP can complete the IPQ-R HP after the patient's visit because it is required that the HP has an idea about the patient's physical and psychosocial condition. Hereafter, these two instruments can be compared easily by the HP. Every HP also needs instructions before using this questionnaire.

3. Implications for clinical practice and healthcare policy recommendations

The results conceived from this PhD project have implications for clinical practice and healthcare policy.

Clinical practice

In clinical practice, HPs and doctors-to-be are not always aware of the commonalities and differences between their own perceptions and the perceptions of patients. They are also not aware that these discrepancies can create problems and difficulties in the patient-provider interaction such as doctor

shopping, miscommunication or mistrust. So, awareness of the importance of illness perceptions by HPs is the first step. The second step is assessing illness perceptions of patients in daily care in order to have knowledge about their perceptions. Not only assessing but also using the CSM as a basis in their communication with their patients. This will give a fast and increased insight in the perceptions of the patient that can be compared immediately with available knowledge of the medical condition or the HP's own representation. The third step is that in the design and construction of clinical interventions, HPs need to address illness perceptions but also the determinants of illness perceptions. The results of the cross-lagged analysis between illness perceptions and health outcomes (Chapter 6) show that in daily clinical practice, beside tackling maladaptive perceptions with for instance cognitive behavioral therapy [25] also a routine screening of depressive feelings and patients' feelings of anxiety is needed. Currently, this is not systematically the case also not in our rheumatology department. The nurse specialist can integrate this in her history taking, make it discussable during interdisciplinary team meetings and ultimately a patient could be referred to a psychologist or psychiatrist. Hereafter, psychological interventions such as specific counseling techniques could be started in order to tackle anxiety and depressive feelings.

Nurses can be trained to give psychoeducation and psychotherapeutic interventions in order to reduce anxiety and depressive feelings. Psychoeducation and psychoeducational interventions cover a broad range of educational activities in combination with counseling and support. Education includes information on treatments, symptoms, training to respond to disease-related problems, and problem-solving strategies to cope with disabilities due to systemic diseases etc. Psychotherapeutic interventions are for instance cognitive-behavioral interventions designed to help patients identify negative or unhelpful thoughts, beliefs, and behaviors, establishing goals to change them, and developing skills to implement helpful behaviors [26]

As mentioned before illness perceptions are significantly associated with various physical, mental and emotional outcomes. When interventions are developed in order to modify illness perceptions, than this might improve patient outcomes. In literature, several interventions are described to tackle illness perceptions, but rare in systemic diseases. For instance, Goodman et al. (2005) [25] found in SLE patients that Cognitive Behavioral Therapy (CBT) changed participants' perceptions of treatment control and emotional representations, and that perceived stress was reduced following the intervention. Chilcot and Moss-Morris (2013) [27] described that in irritable bowel disease, CBT self-management enhanced perceived control over irritable bowel disease, facilitated a more coherent understanding of the illness, and reduced perceptions of the severe and distressing consequences of irritable bowel disease (Chilcot and Moss-Morris, 2013). The CBT based intervention altered illness perceptions which mediated the treatment effects. Also, Broadbent et al. (2009) [28] and Petrie et al. (2002) [29] found that a brief illness perceptions modification programme

in patients with myocardial infarction -which consisted of a patients' personalized education and action plan based on the self-regulation framework- improved patients' perceptions of illness coherence, consequences, timeline and causes ánd could increase the likelihood of their return to usual activities and work.

So, the results described in Chapter 6 show that in clinical practice and in a holistic care approach not only an evaluation of the patients' illness perceptions is needed but also screening of physical and psychological outcomes. Also, in Chapter 2, we found that they were the most influential correlates of illness perceptions and important to take into account when designing interventions.

The results of this PhD thesis show indirectly that in the courses for HPs and doctors-to-be, the concept of illness representations should be mentioned and explained more systematically in addition to the relevance of illness perceptions to understand patients' behavior. Insights about illness perceptions will help HPs not only in understanding their patients' health behavior but also in how they need to give medical information to their patients. They need to know that illness perceptions are less responsive to medical information that is inconsistent with prior or current beliefs or sensations [23] and that if they give information based on the CSM during the medical encounter, the better their perceptions will match with patients' perceptions [30]. Without insight into patients' common-sense representations, medical recommendations often lack relevance and/or context for patients [31].

The results of this PhD study are not only important for the education of physicians but also for the education of nurses. There is a need for more teaching about the importance of illness perceptions of patients and the impact of illness perceptions on patient reported outcomes. When nurses-to-be have knowledge and insight in illness perceptions, its conceptual model and advantages of using this model in daily clinical practice, than they will perhaps be able to unravel and understand maladaptive perceptions of patients and also unexpected health behavior of patients. Nurses and nurses-to-be should be taught to listen to the patients' story, to analyze patients' perceptions and to ask questions directed at gaining more insight in patients' perceptions and tackling maladaptive perceptions. This is possible when they have knowledge about the concept of illness perceptions and practice, for instance in workshops, designing patient education plans and incorporating specific questions about patients' perceptions as the starting point of the education session. Eventually, when they are familiar with the concept of illness perceptions they will be motivated to have attention for it and for its use in routine care. Nurses can educate patients better when they can rely on a model that guides the nurse in different steps until they eventually come to change. For instance, the representational approach to patient education [32] has been developed by nurses and postulates that effective patient education occurs when a nurse starts from patients' knowledge and beliefs before providing new information. Patients' beliefs about their illness are viewed as critical gateways to

change. The model consists of seven key elements and goals in a linear stepwise fashion. This approach can be seen as an interview and is adequately flexible to guide interventions for many different patient care situations.

Healthcare policy recommendations

We found in Chapter 4, that there are different physicians' profiles, that physicians' perceptions about SLE and SSc can be clustered, and that there are divergent illness perceptions among physicians of the same and other disciplines. This poses a challenge in organizing care by several physicians for patients with systemic diseases which are less prevalent and complex. Medical specialists who care for patients with complex multisystem diseases need to have concentrated knowledge concerning all aspects of the disease and resources. The care for these entities should be organized in that way that early diagnosis, patient management and care delivery will be improved [33]. Currently, in Belgium appropriate communication between all care providers of a certain complex patient case is poor. These patients could benefit from treatment in expert or reference centers which are specialized in the care for patients with rare diseases and where integrated interdisciplinary care is developed and guided. Organization and financing of such integrated care is mandatory and deserves specific attention for policy makers on top of facilitating classical treatment innovations that are mainly pharmacological based or introduce new and expensive devices. In Belgium, a start of organizing the care for rare and complex diseases was made by the national government in 2014 (Royal Decree of 25 April 2014 published 8 August 2014 in the Belgian Official Journal/Belgisch Staatsblad) defining the functioning of rare disease networks [34]. Currently, the implementation of all this is still ongoing.

4. New horizons for research in the area of illness perceptions

The results of this PhD are a stepping stone for further research in different areas concerning the topic of illness perceptions. In SLE and SSc, there is a need for longitudinal research to investigate the changes of illness perceptions and their impact on outcomes over time. In this longitudinal model all illness perception dimensions should be implemented together with an insertion of illness identity and causal attributions. In addition to research that uncovers in a prospective way the relationship between illness perceptions and health outcomes, also research in other areas is mandatory as discussed below.

Research in providers' illness perceptions

Qualitative research methods provide a complete and comprehensive understanding when little is known or no research has been done previously [35]. The added value of qualitative research for the studies conducted in this PhD project would be that certain illness perception dimensions that are scored totally differently between groups of physicians or in comparison with patients, can be explored in detail in order to understand physicians' perceptions better. Physicians can express their experience in their own words without being limited by researcher-selected response categories.

In Chapter 4, we found different illness perception profiles of physicians who are part of the multidisciplinary team that takes decisions concerning the therapy and long-term approach of patients with SLE and SSc. It could be interesting to have focus groups of physicians who belong to the same cluster and to explore their beliefs and the contributing factors for those specific beliefs in-depth. Another question that emerges is if these diverse perceptions concerning the condition of the same patient, can affect shared decision making in teams. It is interesting to investigate illness perceptions in multidisciplinary teams but also in interdisciplinary teams, between HPs from different disciplines. The reasons for these differences in perceptions can be explored with qualitative research asking the HPs why they think in a certain way. They could become aware that their education or their own personal models of illness contribute to the formation of illness perceptions concerning the condition of their patient in an appropriate or sometimes perhaps in a contra productive way.

In Chapter 5, we described moderate to large differences between rheumatologists and GPs for the dimensions 'timeline acute/chronic' and 'consequences' for SLE, which needs a further qualitative approach in the format of for instance semi-structured interviews regarding the knowledge and the content of illness perceptions.

Studies conducted to study the link between HP-patient disparities in illness perceptions and their impact on outcomes would also be of great value. Within chronic diseases there are few publications describing the commonalities and differences in illness perceptions between patients and their healthcare providers. Also other researchers in the field [1,23] mention that given the large number of potential areas of misunderstanding and miscommunication that exist in health care, it is surprising that not more work has been undertaken in this area. So, our main aim within this PhD study was to collect data about the differences in perceptions between patients and physicians in the field of systemic diseases which is currently lacking; knowing this is a first step before designing the next phase, i.e. measuring the impact of HP-patient disparities in illness perceptions on patient outcomes and the impact of perceptions of healthcare professionals on patient outcomes. For instance, HP-patient disparities in illness perceptions and their impact on outcomes can be investigated in a cross-sectional study where first differences in HPs' IPQ-R scores and patients' IPQ-R scores can be

calculated. Thereafter, a dissimilarity index for each HP-patient dyad can be computed and correlated with the patient outcome. More information on the contribution of the HP's illness conceptualizations to the variance in patients' coping behaviors is needed. Also studying illness perceptions of HPs as possible predictors of change in patients' health behavior is necessary.

Further exploration of the psychometric properties of the IPQ-R HP

The IPQ-R HP can be optimized for use in clinical practice after taking certain steps. First, an in-depth exploration of the face validity by cognitive interviewing techniques, which is also called think-aloud interviews, is needed. This is a technique that has been used previously in the validation process of illness perception measurements [36,37]. In a think-aloud study participants are asked to read each item and verbalize everything they were thinking regarding that item [38]. So, the purpose of this technique is that researchers explore if items are being understood and interpreted in the presumed way. Second, redundant items or items that are overlapping can be removed or revised immediately whereafter a second round of testing can be planned. This will improve the content validity. Also, dimensions with low Cronbach's alpha coefficients can be discarded.

Third, more insight in the factor structure of the IPQ-R HP conducting exploratory factor analysis is also warranted. In this way, information is gathered about items that can be discarded, where after the questionnaire could be modified. Also information concerning the construct validity is needed. This research should be conducted in large cohorts i.e. multicenter studies, not only in large groups of physicians but also in large groups of HPs, i.e. nurses, social workers, physiotherapists, etc.

Uncovering the illness coherence dimension

In Chapter 6, we found that the predominant direction of effects goes from health outcomes to the illness coherence dimension. This is applicable for both patient groups, SLE and SSc patients and remarkable because literature [39] states that illness coherence may play an important role in longer term adjustment and the response to symptoms. The reverse is true according to our work meaning that health outcomes predict illness coherence and not vice versa.

Moss-Morris and colleagues (2002) [39] described for the first time in the IPQ-R the term illness coherence as 'a type of meta-cognition reflecting the way in which the patient evaluates the coherence or usefulness of his or her illness representation'. Broadbent (2015) and coworkers [20] mentioned in their meta-analysis that illness coherence can be seen as a summary of how the other perceptions fit together to provide a coherent model rather than being an independent illness perception per se. So, further investigation is needed about the role of illness coherence is in the self-regulation process. Is it just the way a patient understands his condition or a summary of all illness perceptions? And how

come that depressive feelings, anxiety and perceived health status precede the formation of mostly illness coherence over time and predicts less the other illness perception dimensions?

Exploring coping as a part of the self-regulation process in patients with SLE and SSc.

As already mentioned in the introduction of this PhD project, which is based on Leventhal's CSM [40], illness representations generate coping efforts which in turn influence health outcomes. Hagger and Orbell (2003) [2] stated that this relationship represent a mediational model in which coping mediates the link between illness representations and health outcomes. Coping as a mediator in the relationship between illness perceptions and outcomes has not been studied in SLE and SSc. In Chapter 6 we did not report coping behavior neither studied the mediating role of coping in the relation between illness perceptions and outcomes. Further research needs to illuminate this because maybe we can understand the results of Chapter 6 better, namely why only consequences and timeline acute/chronic predict outcomes, when we understand the coping mechanism within this relationship.

In conclusion, this thesis contributes to an important aspect of future care in complex chronic diseases. Different aspects were explored such as modifiable correlates of illness perceptions, the preliminary validation of the IPQ-R HP, and an investigation of the prospective associations between illness perceptions and health outcomes such as depressive feelings, anxiety, perceived health status and disease activity. The results of this thesis guide HPs in the selection of factors to take into account when planning interventions for patients. We found that not only emphasis on illness perceptions but on the 'broader picture', the physical and mental functioning is important when striving for a holistic care approach. Zooming into the IPQ-R HP shows that in the future, improvement of the IPQ-R HP is needed but also and most importantly implementing this instrument in standard care and daily practice. Such as for instance how to use the IPQ-R HP in practice and what it will contribute to outcomes. This thesis provides also evidence to what has been experienced in practice by clinicians, namely that physicians from the same and different medical specialties can have common or divergent beliefs regarding the condition of the patient. More studies are needed in the areas of precipitating factors of these divergent beliefs and the impact on interdisciplinary collaboration and decision making.

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PERSONAL CONTRIBUTION

CONFLICTS OF INTEREST STATEMENT

Scientific acknowledgement

In **Chapter 2**, we identified the modifiable correlates of illness perceptions using a systematic review as design. We are very grateful to the librarian of the Biomedical Library, Ms. Marleen Michels, who helped us with the set-up of the search strategy for the different scientific databases.

In **Chapter 3**, we conducted a preliminary validation of the IPQ-R HP. We are very grateful to the doctors, head nurses and nurses from different hospitals and nursing wards for their participation in the validation process of the IPQ-R HP.

In **Chapter 4**, the perceptions of physicians from nine different medical specialties working at three hospitals were identified. We thank all physicians who participated in this study and took their time for completing questionnaires. We are also grateful to the secretaries of the 9 medical disciplines at the University Hospitals Leuven for their help in identifying the doctors. Also, many thanks to Prof. Ben Van Calster, statistician, for conducting the cluster analysis.

In **Chapter 5**, the perceptions of rheumatologists, general practitioners and their patients were analyzed. We want to thank all rheumatologists, general practitioners and SLE and SSc patients for their participation in this study.

In **Chapter 6**, we studied the prospective associations between illness perceptions and health outcomes. We want to thank all SLE and SSc patients for their participation.

Personal contribution

Chapter 2: almost the full work was done by myself after advice from a biomedical librarian.

Chapter 3: I conceived the study together with my supervisors, collected data in collaboration with Master students under my supervision. Analysis, interpretation and writing was done in collaboration.

Chapter 4: I conceived and conducted the full study with my supervisors. Analysis, interpretation of the findings and writing was done together with the statistician and my supervisors.

Chapter 5: I conceived and conducted the full study with my supervisors. Statistical technique was discussed in group and analysis was done by myself with statistical software. Analysis, interpretation and writing was done together with supervisors.

Chapter 6: Idem chapter 5. Statistical analysis was conducted by 2 co-authors, J. Rassart and K. Luyckx. Analysis, interpretation and writing was done together with all co-authors of this study.

Conflicts of interest

There were no conflicts of interests. The studies described in **Chapter 4, 5 and 6** were funded partially by the Fonds voor Wetenschappelijk Reuma onderzoek / Fund for Scientific Research in Rheumatology.

SUMMARY

Illness perceptions are mental models that individuals construct when facing an illness in order to make sense of their symptoms and medical conditions. These illness models can be very specific to the individual which means that patients with the same condition can have different perceptions regarding their illness. Nevertheless, consistent patterns are identified in the way individuals generate illness perceptions. The concept of illness perceptions has been introduced in the early 1980s by Leventhal and colleagues in the Common-Sense Model. This model posits that internal stimuli (e.g. symptom experience such as pain) and external stimuli (e.g. disease-related information from family or healthcare professionals) generate cognitive and emotional representations which guide the selection of coping procedures in order to eliminate and control potential or ongoing 'illness threats'. Afterwards, there is an appraisal of these coping procedures in regulating physical and emotional outcomes.

Literature, which is limited, states that physicians' perceptions about illness are possible correlates of patients' illness perceptions. No clear overview is available in the literature about these correlates of illness perceptions or about the perceptions of physicians or healthcare professionals regarding the illness of the patient. Also, some of these correlates can be determinants or outcomes of illness perceptions. For instance, in the case of anxiety: do patients view their illness more negatively because they are anxious, or do they become anxious because they consider their illness so severe and feel they have no control over it?

This PhD project covers studies addressing the unknowns in literature in patients with systemic lupus erythematosus (SLE) and systemic sclerosis (SSc). Firstly, we identified variables that are associated with illness perceptions. We selected the modifiable correlates of illness perceptions because these variables are prone to change which means that they are modifiable with clinical interventions. We found factors such as medication beliefs, quality of care, illness-related factors, psychosocial factors and information provision and satisfaction. This overview is interesting because healthcare professionals can take this into account in their encounter with patients during patient education or counseling sessions.

Secondly, in order to gather more information regarding physicians' perceptions about SLE and SSc, we set up three studies. In the first study, we performed a preliminary validation of an instrument aimed to measure illness perceptions in healthcare professionals. The second study was a vignette-based study in which we investigated perceptions of physicians from different medical specialties, about SLE and SSc. We found diverse physician profiles based on their perceptions of SLE and SSc which was independent of the frequency of patient contact. In the third study, we examined perceptions of rheumatologists and general practitioners (GPs) about their own patients, this means patients they treat and care for. In the case of SSc, there were more commonalities in perceptions between rheumatologists and GPs and in the case of SLE more differences in illness perceptions.

Thirdly, in the last study we analyzed the link between illness perceptions of SLE and SSc patients and health outcomes such as depressive feelings, anxiety, perceived health status and disease activity spanning one year. We investigated whether illness perceptions are predicted by health outcomes or vice versa. This study was important because almost all literature in illness perceptions in SLE and SSc has a cross-sectional design and assumes that illness perceptions predict health outcomes. In other words, that the direction of the associations goes from illness perceptions to health outcomes. The results of the latter study were that health outcomes such as depressive feelings, anxiety, perceived health status determine illness perceptions one year later. For SLE, we found that illness perceptions are predictors of perceived health status and depressive symptomatology over time.

In sum, this PhD study gave more insight in the modifiable correlates of illness perceptions, the illness perceptions of physicians from various medical specialties and their patients, and uncovered the links between illness perceptions and health outcomes in patients with SLE and SSc. Now, we have more evidence for planning educational interventions for healthcare professionals and for designing clinical interventions for patients but additional research is necessary. Future research is needed on different aspects such as the further investigation of the psychometric properties of the illness perception questionnaire for health care professionals, an evaluation of the impact of divergent illness perceptions of physicians on the organization of care and an investigation of the impact of physicians' illness perceptions on patient outcomes.

SAMENVATTING

Ziektepercepties zijn mentale constructies of modellen die mensen vormen wanneer ze geconfronteerd worden met een ziekte om zo betekenis te geven aan die ziekte of ervaren klachten. Deze mentale modellen kunnen zeer specifiek en individueel verschillend zijn. Dit wil zeggen dat patiënten met dezelfde aandoening verschillende percepties over hun ziekte kunnen hebben. Desondanks bestaat er een consistent patroon in het ontstaan van ziektepercepties. Leventhal en collega's hebben begin jaren '80, voor de eerste maal het concept 'ziekteperceptie' geïntroduceerd en beschreven in het Common Sense Model. Dit model beschrijft hoe interne prikkels zoals bijvoorbeeld pijn en externe prikkels zoals informatie van familie of professionele hulpverleners, cognitieve en emotionele percepties over de ziekte doen ontstaan. De volgende stap in dit model is dat het individu op zoek gaat naar manieren om het hoofd te bieden aan de ziekte om zo de ziekte te elimineren of te controleren. Achteraf zullen deze inspanningen beoordeeld en geëvalueerd worden op vlak van afname van de klachten of verbetering van de ziekte.

Op basis van beperkte literatuur weten we dat de manier waarop een arts kijkt naar de aandoening van de patiënt een bepalende factor is voor de ziekteperceptie van de patiënt. Op dit moment bestaat er geen helder overzicht in de literatuur over deze factoren die gerelateerd zijn aan ziektepercepties van patiënten. Ook is er geen informatie beschikbaar over wat nu precies de percepties zijn van artsen omtrent de ziekte van een patiënt. Daarbij komt nog dat sommige van deze gerelateerde factoren voorspellers of resultaten van ziektepercepties kunnen zijn. Bijvoorbeeld in het geval van angst, bekijken patiënten hun aandoening meer negatief omdat ze angstig zijn of zijn ze angstig omdat ze hun aandoening als ernstig ervaren en het gevoel hebben er geen controle over te hebben?

Dit doctoraatsproject omvat studies die de leemtes opvullen in de literatuur bij patiënten met Systeemlupus (SLE) en Systeemsclerose (SSc). Ten eerste onderzochten we de factoren die geassocieerd zijn met ziektepercepties. We selecteerden de modificeerbare of 'wijzigbare' factoren die gerelateerd zijn aan ziekteperceptie omdat deze variabelen kunnen gewijzigd worden door middel van klinische interventies. Wij vonden factoren zoals medicatiepercepties, kwaliteit van zorg, ziekte gerelateerde factoren, psychosociale factoren en informatie overdracht en voldoening van de verkregen informatie. Dit overzicht is interessant en belangrijk omdat professionele hulpverleners hiermee rekening kunnen houden tijdens patiënten informatie en –educatiesessies.

Ten tweede werden er drie studies opgesteld om meer inzicht te verkrijgen in de percepties van artsen omtrent SLE en SSc. De eerste studie was de ontwikkeling en voorbereidende validatie van een meetinstrument ontworpen om ziektepercepties van professionele hulpverleners te meten. In de tweede studie werden gevalsstudies van patiënten met SLE en SSc aangeboden aan artsen die tewerkgesteld zijn in verschillende medische disciplines met als doel deze gevalsstudies te beoordelen op vlak van hun percepties aangaande SLE en SSc. Wij vonden verschillende artsenprofielen terug los

van het aantal patiëntencontacten dat zij hadden. Hierna onderzochten we percepties van reumatologen en huisartsen over hun eigen patiënten, namelijk patiënten die zij zelf kennen en behandelen. Voor SSc vonden we dat er meer overeenkomsten tussen reumatologen en huisartsen waren in vergelijking met SLE. In SLE werden meer verschillen in ziektepercepties tussen reumatologen en huisartsen waargenomen.

Ten derde, in de laatste studie analyseerden we de link tussen ziektepercepties van patiënten met SLE en SSc en gezondheidsgerelateerde resultaten van ziekteperceptie zoals depressieve gevoelens, angst, gepercipieerde gezondheidsstatus en ziekteactiviteit na een periode van 1 jaar. We waren geïnteresseerd in de vraag of ziektepercepties gezondheidsgerelateerde resultaten voorspellen of zelf voorspeld worden door deze gezondheidsgerelateerde factoren. Deze studie was belangrijk omdat veel onderzoek over ziektepercepties bij SLE en SSc, dwarsdoorsnede onderzoek is wat het onmogelijk maakt om de richting van de verbanden na te gaan. Wij vonden dat gezondheidsgerelateerde resultaten zoals depressieve gevoelens, angst, gepercipieerde gezondheidsstatus, ziektepercepties voorspellen een jaar later. Voor SLE vonden we ook dat ziektepercepties voorspellers zijn van gepercipieerde gezondheidsstatus en depressieve gevoelens over de tijd.

Tot slot kunnen we stellen dat dit doctoraatsproject meer inzichten gaf in de modificeerbare factoren die gerelateerd zijn aan ziektepercepties en in ziektepercepties van artsen uit verschillende medische disciplines over patiënten met SLE en SSc. Ook werd de link tussen ziektepercepties en gezondheidsgerelateerde resultaten bij patiënten met SLE en SSc ontrafeld. De resultaten van dit doctoraatsproefschrift biedt op wetenschap gebaseerde gegevens om de opleiding voor professionele hulpverleners meer vorm te geven en om klinische interventies voor patiënten te plannen. Verder onderzoek is noodzakelijk op volgende gebieden: verdere exploratie van de psychometrische eigenschappen van de ziekteperceptie vragenlijst voor professionele hulpverleners, de impact van uiteenlopende ziektepercepties op de organisatie van de zorg en verder onderzoek naar de impact van ziektepercepties van artsen op gezondheidsgerelateerde resultaten bij patiënten.

CURRICULUM VITAE AND PUBLICATIONS

Seher Arat obtained her bachelor's degree in Nursing at University College Leuven-Limburg (2000) and master's degree in Medical-Social Sciences at KU Leuven (2003). Hereafter, she completed the Academic Teacher's Education Program at KU Leuven (2004). From September 2004 on, she worked as a registered nurse at the University Hospitals Leuven in the Department of Intensive Care, Department of Endocrinology and Department of Rheumatology . Two years later, in September 2006, Seher began working as a Clinical Nurse Specialist in Systemic Diseases at the Department of Rheumatology. In March 2013, she got the opportunity to start a PhD project at the KU Leuven with a focus on illness perceptions in patients with multisystem diseases. In the meanwhile, she was also a guest lecturer in rheumatology nursing at the University College Leuven-Limburg and gave several lectures to staff nurses working at the University Hospitals Leuven about the care approach for patients with systemic diseases.

Publications in international peer-reviewed journals

Arat S, Lenaerts J, De Langhe E, Verschueren P, Moons P, Vandenberghe J, Taelman V, Westhovens R. Illness representations of Systemic Lupus Erythematosus and Systemic Sclerosis: a comparison of patients, their rheumatologists and their general practitioners. Lupus Science & Medicine 2017 (in press).

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D ANKWOORD

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